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RESEARCH ARTICLE

FNAC diagnosis of adenoid cystic carcinoma of the breast in a young female: A rare case report with review of literature

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Manuscript Info Abstract Manuscript History: Adenoid cystic carcinoma (ACC) of the breast although rare and unusual, Received: 29 June 2014 Final Accepted: 29 July 2014

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can be very emphatically diagnosed on fine needle aspiration cytology (FNAC) by applying correct cytological principles. A pathologist should be aware of its benign and malignant mimickers so that a correct diagnosis is rendered in all such cases. This is important because ACC of the breast has an excellent prognosis with rare lymph node and distant metastasis. Biologically also, it behaves differently from other breast malignancies because ACC of the breast are hormonally nonreactive (ER/PR negative). Hence a simple FNAC procedure can hasten the diagnosis and can lead to reduced morbidity (decreased duration of hospitalization and complications associated with biopsy procedure) and also avoid radical mastectomy with axillary lymph node clearance in cases of ACC of the breast.

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Introduction

Adenoid cystic carcinoma (ACC) of the breast is a rare and an unusual neoplastic lesion with a reported incidence rate of 0.1% in literature. [1] Apart from breast, it is also noted in extra mammary sites such as sub mucosal bronchial glands, salivary glands, uterine cervix, bartholin gland and skin. It is however note-worthy that ACC of the breast recapitulates the similar morphology in both mammary as well as extra mammary sites. [2] On cytology, ACC of the breast has to be distinguished from the other benign and malignant conditions which can also have similar cytological pattern/morphology and common shared cellular components. This was more so important in the present case of a young unmarried girl 23 years of age who was diagnosed with ACC left breast on the basis of cytology.

Case Report

A 23 year old unmarried female presented to the surgical outpatient department with complaint of lump in the left breast for the past two months. The patient also complained of tenderness and pain associated with the lump. On physical examination, the breast showed a well circumscribed nodular lump 4x2 cm in size, firm in consistency, tender on palpation, located in the upper lateral quadrant which was adherent to the overlying skin with retraction of the nipple. Mammography of the lump suggested a neoplastic lesion measuring 34x20 mm with irregular speculated margins and distortion of the adjacent parenchyma with no obvious calcification. Fine needle aspiration cytology (FNAC) of the lesion was recommended which was done with a 22 g needle and both air dried as well as alcohol fixed smears were prepared and stained by May Grunewald Geimsa (MGG) and Hematoxylin & Eosin(H&E) stains respectively.

The smears prepared were cellular with monotonous appearing round to oval cells arranged in groups, clusters, in vague microcytic pattern and forming cup shaped tissue fragments as well.[Figure/Table 1A&B] A very characteristic finding noted in the smears examined was metachromatically colored round to cylindrical bodies (hyaline bodies). The material was also noted incorporated with in the three dimensional cell clusters and groups.[Figure/Table 1C & D] The individual cells although appearing bland cytologically, showed crowding, overlapping, definite mild nuclear atypia and coarsening of chromatin. Some of the cells also showed single small

nucleolus. No bare bipolar nuclei or increased myoepithelial cells were noted in the smears examined. Based on these findings, a diagnosis of adenoid cystic carcinoma (ACC) of the breast was made.

In view of a comprehensive cytology report and a suggestive mammography report of malignancy, a tru-cut biopsy was planned. 3 linear cores were obtained which were processed routinely. The sections were taken and stained with H&E stain which showed tumor cells arranged in typical fenestrated nests (cribriform pattern) composed of dual population of cells mainly basaloid cells and few eosinophilic cells. [Figure/Table 2] The stromal spaces at places were filled by hyaline collagen spherules/cylinders. [Figure/Table 3] The basaloid cells had round to ovoid nucleus, scanty cytoplasm with 1-2 nucleoli. Evidence of peri-neural invasion was also documented at few places. [Figure/Table-4] Hence a final diagnosis of ACC of the breast was rendered which was in concordance with cytological opinion. ER/PR/Her2neu stains were negative in the biopsy cores. Subsequently the patient was referred to the oncology department of a higher center for further management.

Discussion

ACC of the breast (Cylindroma - breast) was first diagnosed on FNAC in 1991 by Dong Wha Lee.[3] On FNAC, its distinctive cytological appearance of cribriform pattern and hyaline globules are shared by both mammary as well as extra mammary lesions. Unlike the ACC of extra mammary location, ACC of the breast has a better prognosis and a milder course with rarer incidence of associated axillary lymphadenopathy and distant metastasis. [4] Just like the present case, most of the cases reported have been unilateral at presentation. In literature, ACC of the breast has been reported in a wide age group ranging from 33 year to 97 years but has not been reported in female less than 30 years (23 year in the present case).

Tenderness and pain attributed to the phenomenon of peri-neural invasion in extra mammary ACC has not been much documented in ACC of the reported cases of the breast. [4] In our case, however; peri-neural invasion was noted and this was corroborated with the clinical presentation of tenderness in the lump.

It is imperative that a quick and conclusive diagnosis be rendered on FNAC as this avoids unnecessary surgical biopsy for conformation of the diagnosis and in turn will lessen the associated morbidity, lowering the need for unnecessary hospitalization and the patient can be referred for immediate therapeutic management in cases of ACC of the breast. It is also notable that ACC of the breast has an excellent prognosis and simple mastectomy is often curative for the patient with only rare grade III ACC breast patients requiring radical mastectomy with axillary lymph node clearance. [3]

To reach a conclusive diagnosis of ACC of the breast, a cytopathologist should be aware of certain benign cytological mimickers such as presence of collagen spherulosis (CS) in various benign breast diseases and adenomyo epithelioma (AME) of the breast which can lead to a misdiagnosis of malignancy as the distinctive basement membrane bodies (hyaline bodies) can also be aspirated in these two aforesaid conditions.[5,6] It is more so important when giving a diagnosis of ACC of the breast in young females (as in present reported case).

CS is an associated finding in benign breast diseases such as ductal hyperplasia and other proliferative breast lesions (intraductal papilloma, duct hyperplasia, atypical ductal hyperplasia and sclerozing adenosis). On FNAC, CS can be differentiated from ACC of the breast as in CS, there is a distinct population of myoepithelial cells, bare bipolar cells and other features of benign neoplasia along with spherules; whereas ACC of the breast is a very cellular lesion having distinctive cell groups with no benign cell population or myoepithelial cells / bare bipolar nuclei. Individually the cells of ACC of the breast appear bland and monotonous but none the less show features of atypicality. [7]

The second cytological masquerader, AME of the breast although may contain metachromatic, basal membrane like globules mimicking the finding of ACC of the breast but distinct cellular (dual cell population of epithelial and spindle cells) with naked bipolar nuclei and absence of necrosis or mitoses will comprehensively rule out a possibility of ACC in all such cases.

It is also worthwhile to note that a misdiagnosis of ACC of the breast can also be rendered on histology as intraductal carcinoma and invasive duct carcinoma with cribriform pattern can resemble ACC of the breast to an inexperienced pathologist. Special stains and IHC help in differentiating these from ACC breast [8]. Alcian blue stains the cysts and PAS positivity is seen with ducts. On IHC, markers specific for ACC of the breast are p63 and c-kit which distinguish it from invasive cribriform carcinoma and ductal carcinoma in situ. [9]

The management options in ACC of the breast are divided on the basis of Ro classification with lesions graded from grade I to grade III on the basis of presence of solid component with grade I having mainly glandular and cystic component (in the present case), grade II less than 30% solid component and grade III having more than 30% solid component. Most of the diagnosed cases are of grade I cytology where a simple mastectomy (most preferred) to lumpectomy followed by radiotherapy (alternate method) is done. As the tumor is ER and PR negative, hormonal and chemotherapeutics have not been studied in details. [10] All grade III tumors behave like a conventional high grade ductal carcinoma and treated like so.

Conclusion

The case is worth reporting as it not only documents a case of ACC of the breast in a young 23 year old female but also highlights the importance of correct diagnosis of the entity on cytology alone and importance of differentiating it from its cytological mimickers. This is very essential as simple surgical mastectomy can lead to complete clearance of the cancer in such patients. This in the long run can be beneficial for the patient by reducing not only the morbidity but also reducing the cost incurred by the patient on hospitalization.

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Legends to Figures

Figure/Table 1

Figure/Table 1A: Three dimensional cellular clusters showing metachromatically coloured round bodies (hyaline bodies) [MGGx100]

Figure/Table 1B: High power view showing characteristic "cupping" of the tumour cells around hyaline body [MGGx400]

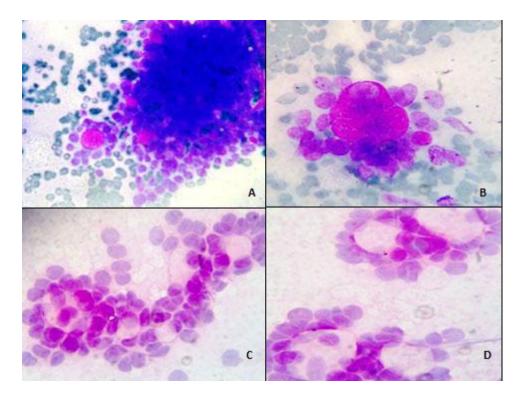
Figure/Table 1C: Presence of monotonous looking hyperchromatic cells encompassing the hyaline bodies [H&Ex200]

Figure/Table 1D: Higher magnification of the Figure/Table 1C [H&Ex400]

Figure/Table 2: A linear core demonstrating tumour islands in characteristic "cribriform pattern" [H&Ex100]

Figure/Table 3: Higher magnification of a tumour island showing monotonous appearing tumour cells tending to form cribriform pattern and showing cystic spaces filled with hyaline bodies [H&Ex200]

Figure/Table 4: Microphotograph depicting presence of tumour island encroaching upon the nerve fibres (perineural invasion) [H&Ex400]



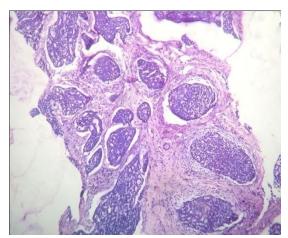
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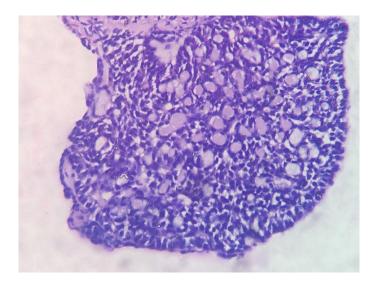
Figure/Table 1B: High power view showing characteristic "cupping" of the tumour cells around hyaline body [MGGx400]

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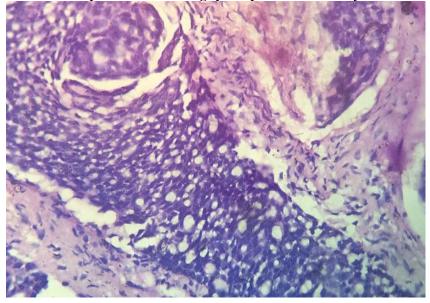
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