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Primary Adenoid Cystic Carcinoma Of The Breast: Case Report And Review Of The Literature

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Abstract

Adenoid cystic carcinoma of the breast is a very rare neoplasm. We report a case of adenoid cystic carcinoma of the right breast presented with painless lump in the upper outer quadrant managed with lumpectomy, axillary lymph node staging and adjuvant local external radiotherapy to the whole breast with simultaneous integrated boost to the site of primary disease using respiratory gated intensity modulated radiotherapy. The available literature is reviewed.

Keywords:

Adenoid cystic cancer breast, mastectomy, adjuvant radiotherapy

Introduction

Adenoid cystic carcinoma of the breast is a very rare neoplasm accounting for 0.1% of all breast cancers [1, 2]. The incidence of regional lymph node metastasis is rare and distant metastasis is uncommon [3, 4, 5]. However distant metastasis has been reported without prior lymph node involvement [1, 6]. Adenoid cystic carcinoma of the breast can occur between 30 to 90 years of age but is more common in woman in the fifth and sixth decade of life [7]. These tumors have excellent prognosis in contrast to extra mammary adenoid cystic carcinoma [1]. Generally these tumors are estrogen (ER) and progesterone (PR) receptors negative [1, 8] & HER-2 negative [9].

Case Report

A 39 year old premenopausal single female presented with two years history of a slowly growing painless small lump in the upper outer quadrant of the right breast. There was no history of nipple discharge and her review of systems was negative for any symptoms. She attained menarche at the age of 14 years. She has no history of smoking and alcohol. Her past medical history was significant for sickle cell trait for which she was not on any medication. She had a significant family history of breast cancer as her maternal cousin was diagnosed with breast cancer at the age of 41 years.

An excisional biopsy showed adenoid cystic carcinoma of the breast. The lesion was measuring 3x2x1.5cm in size with close margins (less than 1mm). There was neither perineural nor lymphovascular invasion identified.

This tumor exhibited solid (<30%), trabecular and cribriform groups with biphasic cell population predominated by small hyper chromatic basaloid cells with pseudo cysts containing eosinophilic material. (Fig. 1). Immunohistochemistry showed the tumor cells to be ER negative with minimal PR positivity. HER-2 was negative. The tumor showed Vimentin positivity (Fig. 2) and focal positivity for S100, Smooth muscle actin and P63 (Fig. 3). This pattern is in keeping with adenoid cystic carcinoma rather than invasive cribriform carcinoma as in the latter the tumor is invariably ER and PR positive. The C-kit on this lesion showed very faint and focal positivity (Fig 4).

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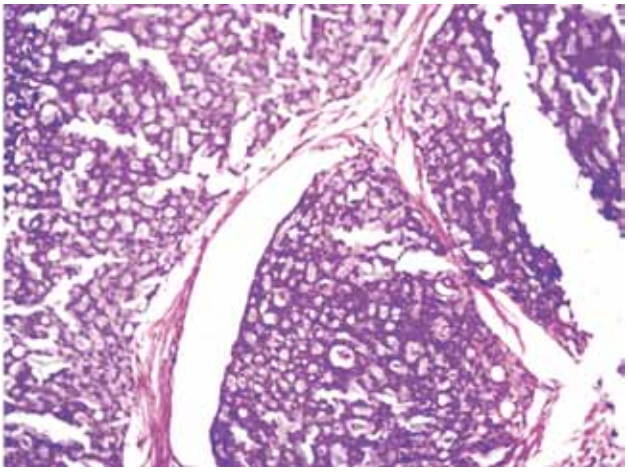

Figure 1: (H&E Medium Power X 200) showing growth of tumor with cribriform pattern.

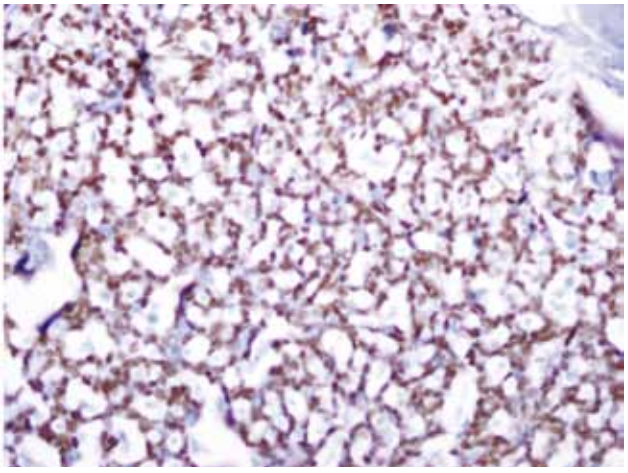

Figure 3: (High Power X 400) tumor cells show P63 nuclear positivity by Immunohistochemistry.

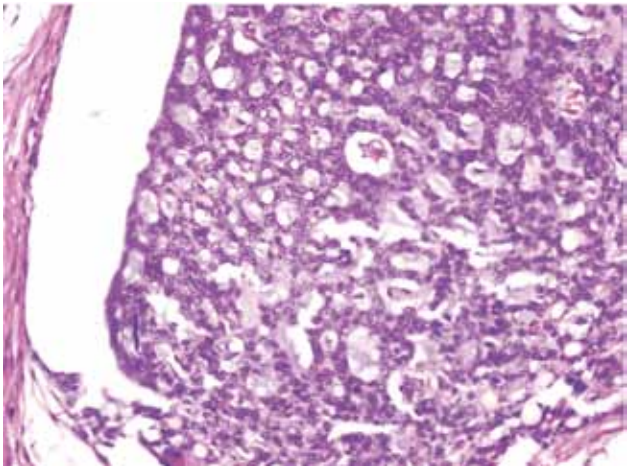

Figure 2: (High Power X 400) showing tumor cells with vimentin positivity by immunohistochemistry.

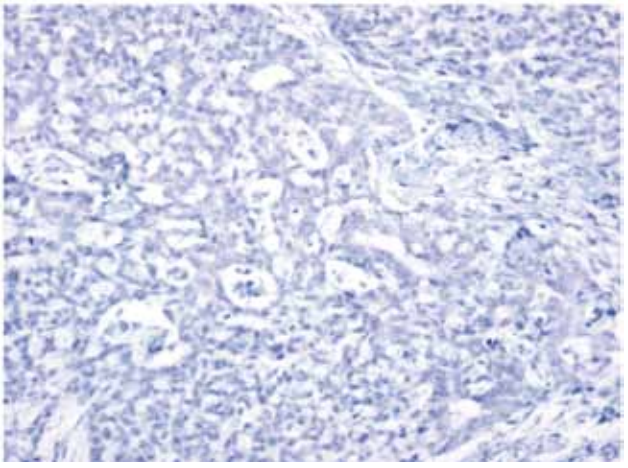

Figure 4: (High Power X 400) C-kit, focal and faint positivity in high power

Post lumpectomy breast ultrasound showed distorted architecture in the upper outer quadrant of the right breast at the site of previous surgery representing postoperative changes. No obvious mass lesion was seen in either breast. Both axillae showed benign looking axillary lymph nodes. The staging CT scan of the chest, abdomen and pelvis did not show any metastatic disease. Ca 15.3 was in the normal range 18.4 (ref 0 - 31.3). Two weeks later, she underwent wide local excision and sentinel lymph node biopsy. The pathology showed fibrocystic changes involving the remaining breast and one mm focus of atypical ductal hyperplasia. There was no evidence of residual malignancy. The two lymph nodes dissected were negative for the malignancy. All excisional margins including skin were clear. She was staged as T2N0M0 adenoid cystic carcinoma of the right breast. She subsequently received adjuvant local external radiotherapy using respiratory gated intensity modulated radiotherapy (IMRT) technique to a dose of 45 Gy in 25 fractions to whole of the right breast with concomitant boost to the tumor bed to a total dose of 60 Gy in 25 fractions.

Discussion

Adenoid cystic carcinoma of the breast is a rare and slow growing tumor accounting for 0.1% of all breast neoplasm with approximately one case occurring per 1 million female per year [1,4,9]. To our knowledge, largest series describing adenoid cystic carcinoma of breast is from USA describing 300 cases in their study [9]. In contrast to the aggressive nature of adenoid cystic carcinoma that occurs in the head and neck region, adenoid cystic carcinoma of the breast has a very favorable prognosis[1]. The most frequent

presenting symptom is tender breast mass [1, 9] which was absent in our patient. The local pain in adenoid cystic carcinoma of the salivary gland is believed to be due to perinural invasion which is rare in adenoid cystic carcinoma of the breast [1]. The review of literature shows that radiological appearance are generally non-specific and it can present as a benign appearing, smooth, round or lobulated density or as an irregular mass[1]. The mammography shows a circumscribed lobulated nodule, usually in the upper outer quadrant or in a peri-areolar location [10]. The majority of adenoid cystic carcinoma of the breast is ER –negative /PR –negative, however hormone receptor positive tumors have also been described in minority of cases [9]. HER-2 neu – negative status is seen in these tumors [9]. The vast majority of adenoid cystic carcinoma of the breast shows positivity for c-Kit particularly the subtype with cribriform morphology [11, 12, 13]. This is in keeping with the results of our case.

Pathologically, various growth patterns have been described in adenoid cystic carcinoma of the breast as glandular (cribriform), tubular and solid (basaloid) types [1]. The adenoid cystic carcinoma of the breast is graded according to the solid proportions of the tumor which in turn correlates with the prognosis [14]. Grade 1 (No solid element), Grade 2 (<30% solid element) and Grade 3 (>30% solid elements). Tumors with a higher proportion of solid elements tend to be larger with a higher risk of recurrence and have a more aggressive clinical course [10, 14]. The molecular marker study by Arpino et al found 92% of patient were DNA diploid in adenoid cystic carcinoma of the breast whereas DNA aneuploidy was noted in approximately 60-65% of patient in invasive breast cancer[7]. In contrast to triple negative breast cancer, adenoid cystic carcinoma of the breast rarely involved regional lymph nodes and are associated with excellent prognosis[9].

There is no consensus on optimal treatment management for patients with adenoid cystic carcinoma of the breast as this is a rare diagnosis [1]. Because of lack of randomized controlled trials and sparse literature on this topic, we don't know much about the optimal management of this disease. Reported treatment modalities include simple mastectomy, wide local excision with local radiotherapy and radical mastectomy [1]. Due to low incidence of nodal metastasis routine lymph node dissection is not indicated [15]. High rates of local recurrence have been reported after local excision only [6, 10]. Lemming et al showed that 9 out of 24 patients (37%) had local recurrence after local excision [16]. Single or multiple recurrences may occur years after initial diagnosis [9]. A case report of local recurrence of adenoid cystic carcinoma in the breast after successful treatment with lumpectomy and adjuvant chemotherapy and radiotherapy was described [17]. Ro et al proposed local excision for grade 1 tumors, simple mastectomy for grade II tumors and mastectomy with axillary node dissection for grade III tumors [10]. The role of radiotherapy, chemotherapy or hormonal therapy is unproven[6]. Synchronous and metachronous in situ and invasive carcinomas have been described in the ipsilateral breast as well as in the contralateral breast in patients with adenoid cystic carcinoma of the breast and the prognosis will be of the other histological subtype[9]. Distant metastasis usually to lung or other organs can occur without positive axillary nodes [18].

Conclusion

Adenoid cystic carcinoma of the breast requires accurate diagnosis as this is a rare entity. The prognosis is excellent with long term cure rate. The patient needs close follow up as local recurrence and distant metastasis may occur even after appropriate treatment.

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