Salivary Gland Like Breast Carcinoma/Adenoid Cystic Carcinoma: Case Report

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ABSTRACT

Adenoid cystic carcinoma of the breast constitutes approximately 0.1% of all breast tumors. They can be located in the trachea, bronchus, cervix, lacrimal gland, and skin as well as the breast. Tumors in the breast have better prognoses compared to those in other locations. The diagnosis and treatment planning of this tumor is challenging due to its rare incidence. In this article, we presented a case that was diagnosed with adenoid cystic carcinoma of the breast upon pathology evaluation.

A 59-year-old female patient was admitted to our clinic due to a mass in her right breast. Her mammography revealed a 1 cm in diameter mass in the upper outer quadrant of the right breast, which was classified as BIRADS 4C (Breast Imaging Reporting and Data System). On magnetic resonance imaging (MRI) the lesion was also reported as BIRADS 4C. The patient underwent breast conserving surgery (BCS), and the pathology result was reported as adenoid cystic carcinoma of the breast. The patient received chemo-radiotherapy in the postoperative period.

Adenoid cystic carcinoma of the breast has been first described in the salivary glands. They can be confused with benign lesions both on physical and radiological examinations. Sentinel lymph node biopsy (SLNB) can be used since axillary metastases are rare. Local recurrence and distant metastases are also very rare. Usually, BCS followed by radiotherapy is adequate to obtain local control. In selected patients with a poor prognosis, chemotherapy and hormonal therapy should be added to the treatment.

Key words: Adenoid cystic carcinoma, breast carcinoma, sentinel lymph node biopsy

Introduction

Adenoid cystic carcinoma (ACC) is a malignant breast neoplasm with a very low incidence, constituting only 0.1% of all breast cancers (1). ACC morphologically consists of a combination of proliferative glands (adenoid part) and stromal elements (pseudo-glandular portion) (2). They can be located in the trachea, bronchus, cervix, lacrimal gland, and skin as well as the breast. Tumors in the breast have better prognoses compared to those in other locations. Lymph node involvement and distant metastases are rarely observed (3, 4). Estrogen (ER) and progesterone (PR) receptors are usually negative. The diagnosis and treatment planning of this tumor is challenging due to its rare incidence. In this article, we presented a case that was diagnosed with adenoid cystic carcinoma of the breast upon pathology evaluation, after obtaining informed consent from the patient.

Case Presentation

59 years old female patient was admitted to our clinic due to a mass in her right breast. Her past medical history revealed diabetes mellitus, Hashimoto’s thyroiditis, hemorhoidectomy in 2008 and lumpectomy from the left breast. The pathological examination of the excised mass was reported as fibrocyst. She was using oral anti-diabetic and levo-thyroxine therapy. She did not have family history of breast cancer. She did not smoke or used alcohol. On physical examination, a regular bordered mass was detected in the upper outer quadrant of the right breast. There were no signs of erythema, ecchymosis, skin ulceration or retraction. Her mammography revealed a 1 cm in diameter mass in the upper outer quadrant of the right breast, which was classified as BIRADS 4C. On magnetic resonance imaging (MRI), the lesion was also reported as BIRADS 4C. The patient underwent wire-guided lumpectomy. A 5x4x3.5 cm in size excisional biopsy specimen was sent for pathologic evaluation. The lesion appeared well circumscribed and fibrotic on cross-section. On microscopic evaluation, a focal infiltrative tumor that showed trabecular and tubular pattern within a fibrotic stroma was observed (Figure 1). Neoplastic cells were basoloid type, with small hyperchromatic nuclei and narrow cytoplasm. True glandular structures were also detected. The immunohistochemical studies showed focal positive staining for myoepithelial cell markers CD10 and smooth muscle actin, as well...
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reported patient aged 31 years and the oldest 86 years (8, 9). Our pa-tient’s age was 59 years, in concordance with the literature.

The most common clinical manifestation in patients with adenoid cystic carcinoma is usually a palpable mass located around the areola and pain (8, 10). Our patient also complained of a palpable mass. In rare cases, it may represent in the form of nipple discharge (11). In a retrospective study by Mc Crenath et al., 10 out of 22 patients were admitted with a complaint of breast pain (12). The presence of pain and tenderness in the breast are probably indicators of perineural invasion of the tumor. The tumor can be diagnosed by Hematoxylin eosin (HE) staining. Kal-
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ACC, which is a well-circumscribed, mobile mass, can be confused with benign lesions radiologically. Kasagawa described spicular or well-
circumscribed, lobular, high-density masses that do not form a cluster of microcalcifications on mammography (MMG). On ultrasound (US) lesions appear hypoechoic (13). Similar MMG and US findings have been described in our patient.

Figure 1. The tumor showing trabecular and tubular pattern within fibrotic stroma

as CD 117. Based on these findings, the case was accepted as adenoid cystic carcinoma of the breast. The tumor distance to surgical margins was 1 cm. Therefore, the patient underwent breast-conserving surgery (BCS), and no residual tumor was detected. None of the eight dissected axillary lymph nodes was metastatic. The tumor was T1N0M0, and ER (-), PR (-), Cerb B-2 (-). After the operation, the patient received adjuvant chemo-radiotherapy. The patient is being followed-up for 40 months, without local recurrence and distant metastasis.

Discussion and Conclusions

Salivary gland like carcinomas of the breast include acinic cell carcinoma and adenoid cystic carcinoma. Although mucoepidermoid carcinoma are rare, some authors classify these type of tumors within metaplastic carcinoma (5). Adenoid cystic carcinoma (ACC) is a malignant breast neoplasm with a very low incidence, constituting only 0.1% of all breast cancers (1). Its prognosis is good, and lymph nodes and distant metastases are very rare (3, 4). ACC is more common in Caucasians and women, there are also case reports of male patients (6, 7). They are more common in the 5th and 6th decades, with the youngest reported patient aged 31 years and the oldest 86 years (8, 9). Our patient’s age was 59 years, in concordance with the literature.

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Axillary lymph node metastases are rare in patients with ACC. Sumpio et al. (14) reported axillary lymph node involvement only in one patient in their series of 120 patients. Arpino et al. (15) detected only 4 patients with axillary lymph node metastases in a study of 182 cases. In the same study, distant metastases were found in 14 cases, and they had a tendency for distant metastases without axillary metastases. For that reason, sentinel lymph node biopsy (SLNB) should be considered in patients with ACC rather than axillary dissection, which is a radical procedure. Axillary dissection should be performed if sentinel lymph node sampling reveals metastatic carcinoma. Thus, the morbidity of axillary lymph node dissection is avoided. In our patient, the axillary dissection did not show any metastatic lymph nodes.

Distant organ metastasis is also rare in ACC. The most common metastasis site is the lung (13, 14). Other areas of metastases include the liver, brain and kidneys (12, 16).

Due to the low incidence of ACC, a standard treatment approach has not been defined. Treatment options vary from simple lumpectomy to radical mastectomy. Currently, radical mastectomy is no longer rec-

ommended due to the low rate of axillary metastasis and the physical and psychological damage this method causes in patients. Instead, many surgeons prefer simple mastectomy. On the other hand, simple lumpectomy has the advantages of low operative damage, less post-operative discomfort and faster healing. Nevertheless, simple lumpec-
tomy is not recommended due to high local recurrence. Santamaria et al. reported high local recurrence rates in patients with simple lumpe-
tomy (17). Leeming et al. (18) detected a high local recurrence rate af-
ter lumpectomy of 37%. Youk et al. (19) stated that systemic adjuvant chemotherapy and radiotherapy should be used after simple lumpectomy. With this approach, local recurrence rates may be reduced. Simple mastectomy may be more suitable especially if the patient is difficult to follow-up, or has low socio-cultural or economic level. Considering our patients’ age and socio-cultural level, breast-conserving surgery was applied. We also used adjuvant chemotherapy and radiotherapy. In a 40-month follow-up period, our patient did not develop any local and distant organ metastasis.

Examination of hormone receptor status usually reveals ER and PR negativity in AAC. Leeming et al. (18) identified only one patient with ER positivity in their series of 140 patients. In our patient, the ER, PR and c-erb-B2 receptors were negative; therefore, a triple negative breast cancer was detected. Although triple negative invasive breast cancer is known to have a poor prognosis, patients with ACC have variable outcomes. Previous studies reported patients who received tamoxifen as adjuvant hormonal treatment, but since hormone receptors were negative in our patient, hormone therapy was not used.

In conclusion, SLNB can be used in patients with ACC since axillary metastases are rare. In addition, except those treated with simple lumpectomy, BCS and adjuvant radiotherapy is accepted as adequate treatment due to the low rates of local recurrence and distant metas-
tases. In selected patients with a poor prognosis, chemotherapy and hormonal therapy should be added to the treatment.

Ethics Committee Approval: N/A.

Informed Consent: Written informed consent was obtained from patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - T.A., M.K.A.; Design - T.A., S.C.C.; Su-

Financial Disclosure: The authors declared that this study has received no financial support.

Conflict of Interest: No conflict of interest was declared by the authors.

References