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Male Breast Adenoid Cystic Carcinoma

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Key Words
Male breast cancer · Adenoid cystic carcinoma · Bone metastasis

Abstract
Introduction: Adenoid cystic carcinoma (ACC) of the breast is a rare condition, and cases in male patients are even less common. ***Case:*** We describe a case of ACC of the breast with axillary lymph node metastasis, disseminated osteolytic bone metastasis and bone marrow involvement in a 41-year-old man. ***Conclusion:*** Male breast ACC is an extremely rare malignancy; there can be difficulty in obtaining a final diagnosis. We report this case because of its rarity.

Introduction

Breast cancer in men is very rare, and the majority of male breast cancers show pathological findings similar to breast cancer in females. Guidelines for diagnosis and treatment are based on those for female breast cancer due to the extremely low incidence of male breast cancer, i.e. <1% of the rate in females [1]. Male breast cancer usually presents as a firm, painless mass, along with palpable axillary nodes, nipple retraction and ulceration of the skin at presentation. The mass is usually located in the subareolar region, but can also be seen in the upper outer quadrant [2]. Adenoid cystic carcinoma (ACC) of the breast is a rare subtype of breast cancer, usually originating from the salivary gland. ACC of the breast has very favorable biological characteristics for treatment and patients usually have an excellent prognosis. Good local control can be achieved by lumpectomy with radiation or by simple mastectomy. Axillary lymph node dissection is not helpful for clinical management due to

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the rarity of metastasis [3]. ACC of the breast is a rare subtype of breast cancer, and cases in males are even less common. We report a rare case of advanced ACC of the breast in a 41-year-old man, along with a review of the relevant literature.

Case Report

The patient was a 41-year-old male who presented at the Department of Rehabilitation with symptoms of cervical back pain that had been occurring for 2 months. He was healthy, with an unremarkable medical history and a 20-pack per year smoking history. A cervical spine CT scan showed osteolytic changes with bony fragmentations in the C3, C5 and C6 transverse processes and vertebral bodies. These findings are suggestive of pathological fracture related to the presence of a tumor, such as metastatic carcinoma or multiple myeloma, rather than trauma (fig. 1a). To differentiate between metastatic bone lesions and the presence of a solid tumor, we examined the bone scan labeled with ^{99m}Tc, and the results showed only osteolytic bone lesions, with the exception of a small number of fractures. Therefore, we strongly suspected the presence of multiple myeloma (fig. 1b). We did a serum and urine protein electrophoresis evaluation and immunofixation, but there was no evidence of monoclonal gammopathy. In the bone marrow examination for excluding hematologic malignancies such as multiple myeloma and lymphoma, the findings showed that the hematopoietic cells were almost replaced by neoplastic cells with a glandular pattern. Neoplastic cells were stained with CK PAN (AE1/AE3) and were positive (fig. 2a–f).

Metastatic carcinoma was suspected, but there were no abnormal findings in the esophagogastroduodenoscopy or colonoscopy. A PET-CT scan showed a disseminated hypermetabolic lesion (maxSUV = 7.4) in the axial skeleton, multiple, small-sized lung nodules and a multifocal hypermetabolic lung lesion (maxSUV = 2.6) as well as focal hypermetabolic lesions in the left axillary lymph nodes (maxSUV = 2.3) (fig. 1c). An ultrasound-guided core biopsy of the two 1.7-cm, low-echoic, pathological lymph nodes in the left level I axilla area was performed. The result of core biopsy was ACC (fig. 2g, h). Otorhinolaryngology and head and neck screening were performed to attempt to ascertain the location of the primary tumor in the salivary gland, as most ACC tumors in men are of salivary gland origin. However, this screening failed to identify specific lesions.

To identify the primary origin of the cancer, we reviewed the images and performed a whole-body physical examination. This revealed palpable and movable small nodular lesions in the periareolar area of the left breast, which had escaped interpretation on the PET-CT scan because the lesion was obscured (fig. 1d). There were clustered small nodules at the 1 o'clock and 2 o'clock locations around the left nipple upon physical examination. In a breast ultrasound, an irregularly-shaped solid lesion, 1.7 cm in size, was observed in the left subareolar area. The margin of the lesion was irregular and the internal area was relatively homogeneous and hypoechoic and was suggestive of a BI-RADS category 4C lesion. A core biopsy was performed and the pathology result was ACC, the same as that of the auxiliary lymph node biopsy (fig. 2i, j). Immunohistochemical staining was performed and the sample was negative for estrogen receptor (ER), progesterone receptor (PR) and c-erbB2, and positive for CK5/6 and P53. We finally diagnosed ACC of the breast with multiple lung and bone metastases and bone marrow involvement.

The authors submitted a waiver from their institutional review board (IRB) stating that this case report does not require IRB approval or oversight.

Discussion

The overall incidence of ACC of the breast is 0.05–0.1% [3, 4], and is exceedingly rare in men. ACC commonly occurs at the following sites: the minor or major salivary glands, oral mucosa, larynx, lung and maxillary sinus. The incidence of all types of pathology of breast cancer in men is <1% in Western countries [1], but the exact incidence in Korea is not known due to the rarity of this disease in men.

According to a subsequent review of the relevant literature of ACC of the breast, this cancer generally affects older female patients, with a median age range of 58–64 years [5]. The median age of male breast ACC patients is not clear because few reports exist; however, of the cases published [6–8], 2 patients were diagnosed during adolescence and another at the age of 82 years. In our report, the case of a 41-year-old, relatively young man with a nontender, movable, left-breast lesion is described. ACC of the male breast can develop at a relatively younger age than the common male breast cancer pathological type. The published tumor sizes of male breast ACCs range from 1.8 to 3.7 cm [5], and in this case, the maximum tumor diameter was 1.7 cm.

ACC of the breast is not associated with a bloody nipple discharge, even though it is present near the nipple areola region [9]. Our case also had no subjective symptoms, with the exception of the small palpable nodularity in the periareolar area. One hundred and fifty cases of female ACC of the breast have been reported in the literature, only two of which had axillary lymph node metastasis [10–12]. However, our case showed pathologically confirmed left axillary lymph node metastasis and bone marrow involvement. This finding suggests the possibility of the long-term duration of the disease prior to the diagnosis, due to the small-sized primary tumor and lack of symptoms or signs except for the presence of aggressive multiple bone and lung metastasis.

This case displays an extraordinary pattern of bone metastasis findings compared to other solid tumors: CT and PET-CT scans showed osteolytic bone lesions, but the findings of bone scans labeled with ^{99m}Tc were negative. This reflects almost all lesions as having osteolytic activity without compensatory osteoblastic activity, like in multiple myeloma. This pattern could be a characteristic bone metastasis finding of male breast ACC.

The majority of breast ACCs are triple-negative (i.e. ER-, PR- and HER2-negative), according to previous studies and reports [13]. In this case, negative findings of ER and PR were observed, consistent with reported literature. Male breast ACC is rare, and only a few cases have been reported [6–8]. It is difficult to trace the cancer's origin to the male breast when initial pathological findings reveal ACC in metastatic lymph nodes, as in this case. Salivary-gland origin tumors are suspected first because ACC with this site of origin is common in men. Furthermore, it was difficult to identify the primary site as breast cancer in this case because there was no skin retraction or nipple discharge, unlike common types of breast cancer.

In contrast to triple-negative breast carcinomas, ACC of the breast has an excellent prognosis for female patients, as the incidence of lymph node metastasis is lower and distant metastasis is uncommon. ACC of head or neck origin is also known to have a slow progression and thus a good prognosis for patients. Previously reported cases of male breast ACC also show relatively good clinical courses. However, male breast ACC could be overlooked by the patient, and this, coupled with the difficulty of diagnosis, can lead to extended periods of time, with possible aggressive metastasis, until a diagnosis is made.

References

1 Jemal A, Siegel R, Ward E, Hao Y, Xu J, Thun MJ: Cancer statistics, 2009. CA Cancer J Clin 2009;59:225–249.
2 Giordano SH, Cohen DS, Buzdar AU, Perkins G, Hortobagyi GN: Breast carcinoma in men: a population-based study. Cancer 2004;101:51–57.
3 Arpino G, Clark GM, Mohsin S, Bardou VJ, Elledge RM: Adenoid cystic carcinoma of the breast: molecular markers, treatment, and clinical outcome. Cancer 2002;94:2119–2127.
4 Azzopardi JG, Ahmed A, Millis RR: Problems in breast pathology. Major Probl Pathol 1979;11:i–xvi, 1–466.
5 Ghabach B, Anderson WF, Curtis RE, Huycke MM, Lavigne JA, Dores GM: Adenoid cystic carcinoma of the breast in the United States (1977–2006): a population-based cohort study. Breast Cancer Res 2010;12:R54.
6 Kshirsagar AY, Wader JV, Langade YB, Jadhav KP, Zaware SU, Shekhar N: Adenoid cystic carcinoma of the male breast. Int Surg 2006;91:234–236.
7 Liu J, Jia W, Zeng Y, Deng H, Rao N, Su F: Adolescent male adenoid cystic breast carcinoma. Am Surg 2012;78:E288–E289.
8 Miliauskas JR, Leong AS: Adenoid cystic carcinoma in a juvenile male breast. Pathology 1991;23:298–301.
9 Wells CA, Nicoll S, Ferguson DJ: Adenoid cystic carcinoma of the breast: a case with axillary lymph node metastasis. Histopathology 1986;10:415–424.
10 Sinn HP, Lehnert T, Otto HF: Adenoid cystic cancer of the breast. Case report and meta-analysis of the literature. Chirurg 1993;64:198–202.
11 Qizilbash AH, Patterson MC, Oliveira KF: Adenoid cystic carcinoma of the breast. Light and electron microscopy and a brief review of the literature. Arch Pathol Lab Med 1977;101:302–306.
12 Cavanzo FJ, Taylor HB: Adenoid cystic carcinoma of the breast. An analysis of 21 cases. Cancer 1969;24:740–745.
13 Kontos M, Karles D, Petrou A, Alexandrou PT: Adenoid cystic carcinoma intermingled with ductal carcinoma of the breast: a case report and review of the literature. J Med Case Rep 2011;5:437.

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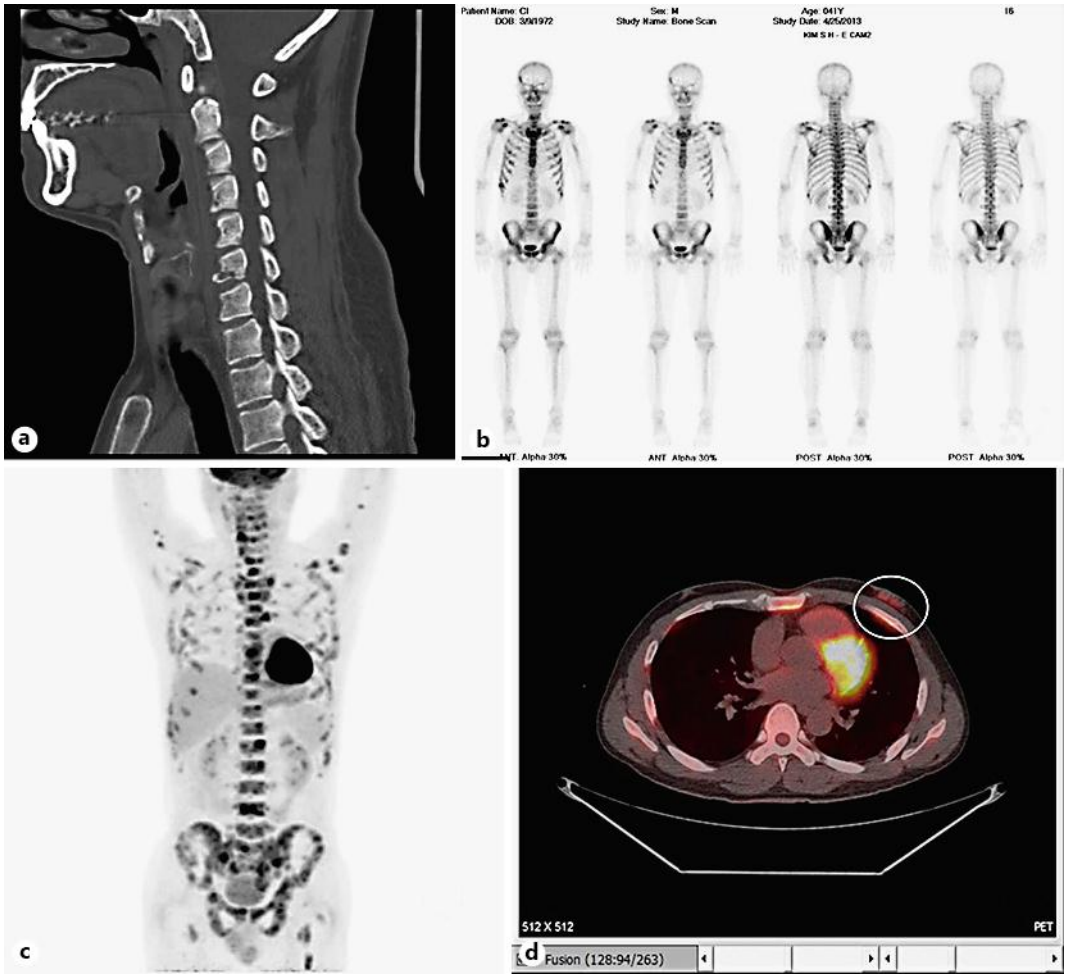

Fig. 1. **a** The cervical spine CT scan showed osteolytic changes with bony fragmentations in the C3, C5 and C6 transverse processes and vertebral bodies. **b** A bone scan showed multiple rib fractures, but otherwise no remarkable uptake. **c** A PET-CT scan showed a multifocal, disseminated hypermetabolic lesion (maxSUV = 7.4) in the axial skeleton with metabolic lesions in the left axillary lymph node (maxSUV = 2.3) and both lungs (maxSUV = 2.6). **d** A PET-CT scan showed a left-breast mass with small daughter nodules (in the white circle) (maxSUV = 1.9).

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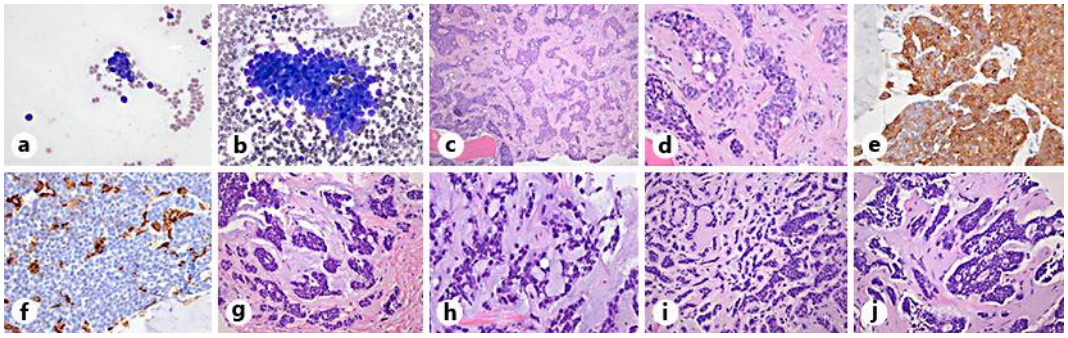

Fig. 2. A bone marrow aspiration and biopsy showed neoplastic cell clusters. **a** BM touch print. Wright. ×400. **b** BM aspiration. Wright. ×400. **c** BM biopsy. HE. ×100. **d** BM biopsy. HE. ×400. **e** BM biopsy. Cytokeratin. ×400. **f** BM biopsy. CD45. ×400. Histopathology of metastatic ACC in the left axillary lymph node (**g**: HE, ×100) and (**h**: HE, ×400). Histopathology of adenoid cystic carcinoma in the left breast (**i**: HE, ×100) and (**j**: HE, ×400).