Invasive Ductal Carcinoma Arising from Ectopic Breast Tissue in Axilla: A Case Report

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Primary carcinoma of ectopic breast tissue is rare. We present a 57-year-old female patient with a large erythematous bulging mass in the right axilla. Imaging studies did not reveal any other lesions except for the one detected in the right axilla. Wide excision with axillary lymph node dissection was performed. The pathology report revealed a poorly differentiated carcinoma with no surrounding lymphoid tissue or lymphovascular infiltration or noncancerous breast tissue. We think that in our case the tumor was probably derived from a primary carcinoma of ectopic breast tissue rather than from a metastatic tumor or occult breast cancer. One year later, the patient presented with a local recurrence at the ipsilateral breast. Mastectomy with chemotherapy was performed. After three cycles of chemotherapy, imaging studies revealed distant metastases. This case report and literature review describe the characteristics of ectopic breast cancer in the axilla and reflects on which initial management strategy is appropriate.

Key Words: Axilla, Breast neoplasms, Human mammary gland

INTRODUCTION

Primary carcinoma of ectopic breast tissue has been reported only in a small number of cases. Ectopic axillary breast tissue differs from the axillary components of the tail of Spence, because it develops as a result of the failed resolution of the mammary ridge, an ectodermal thickening that extends from the axilla to the external genitalia [1]. Copeland and Geschickter [2] proposed a classification of ectopic breast tissue, in which accessory nipple formation, areolar formation, or both, with or without the glandular breast, is termed supernumerary breast. In contrast, an aberrant breast refers to ectopic breast tissue that lacks a nipple or the areolar complex. Thus, we refer to an aberrant breast as ectopic breast tissue.

CASE REPORT

A 57-year-old female patient noticed an erythematous bulging lesion in her right axilla for 2 months. There was also a palpable mass in her left breast that had been present for several years, but had not been evaluated further. Physical examination showed a tender and poorly defined mass measuring 5 cm in diameter with pustular discharge from the central punctum in the right axilla, and a well-defined, round mass in the left breast. There was no accessory nipple or areolar complex in either axilla. The early diagnosis was infected epidermal inclusion cyst or lymphadenitis. To confirm this diagnosis, we performed a fine-needle aspiration (FNA) cytology test, which suggested a probable metastatic carcinoma (Figure 1).

A mammography showed no lesions in the right breast and a well-defined, hyperdense mass in the left breast. Ultrasonography revealed a microlobulated, hypoechoic mass with irregular margins at the right axilla, no mass in the right breast and a well-defined, hypoechoic mass in the left breast. Contrast-enhanced magnetic resonance imaging showed a right axillary mass measuring 8×3.8 cm and conglomerated, enlarged lymph nodes with heterogeneous enhancement and an enhancement kinetic curve resembling a washout pattern. However, there were no abnormal findings in both breasts, except for a benign mass in the left breast. Positron emission tomography-computed tomography (CT) and chest CT showed no other lesion except those mentioned above (Figure 2). Routine hematological and biochemical parameters as well as tumor markers (carcinoembryonic antigen, CA 15-3, CA 125) were within normal ranges. An ultrasound-guided core...
needle biopsy was performed. Pathological examination suggested a poorly differentiated invasive carcinoma of unknown origin.

The surgical treatment of choice was a wide excision of the right axillary mass with axillary lymph node dissection and an excisional biopsy of the left breast mass. Pathological examination revealed a tumor measuring 6×4×3.5 cm abutting the metastatic lymph nodes. There was no surrounding lymphoid tissue or lymphovascular infiltration in the specimen, which was indicative of a primary carcinoma rather than a metastatic carcinoma. There was an irregular subcutaneous proliferation of fibrous connective tissue infiltrated by an invasive ductal carcinoma (scirrhous carcinoma), with a high histological and nuclear grade (Figure 3). Since there were no regions of noncancerous breast tissue and immunohistochemical analysis performed on paraffin sections to detect the estrogen receptor (ER), progesterone receptor (PR), and c-erbB-2 yielded negative results, additional immunostaining procedures were performed to differentiate invasive ductal carcinoma of the breast from other carcinomas including skin adnexal tumors. The tumor cells were negative for the S100 protein and focally positive for cytokeratin 5/6 and cytokeratin 19. The above histological and immunohistochemical findings were suggestive of invasive ductal carcinoma. A total of five out of 16 lymph nodes were positive for tumor infiltration. There was no residual tumor on the cut margin. Pathological examination of the left breast mass revealed a fibroadenoma.

Postoperative treatment included chemotherapy (docetaxel, doxorubicin and cyclophosphamide combination therapy, six cycles) and radiotherapy. One year later, a regional recurrence was found on the parasternal area of the right breast. A total mastectomy was performed and the patient received chemotherapy treatment (paclitaxel monotherapy, three cycles). The pathological reports revealed multiple small-sized, recurrent masses on the inner portion of the right breast. After chemotherapy, follow-up evaluation with a chest CT revealed multiple suspicious metastatic lymphadenitises on the bilateral internal mammary, left axilla and right mediastinum. After the chemotherapy regimen was modified to gemcitabine and vinorelbine, the patient has been in a tolerable state with a partial response.

**DISCUSSION**

The overall incidence of ectopic breast tissue is reported as 6% of the
Among this population, ectopic breast carcinoma is rare and is reported as a disease that shows variations in incidence rate. Chiari [4] reported that ectopic breast carcinoma accounts for 0.3% of all breast cancers in one series, whereas it was 0.2% to 0.6% in Japan. The ectopic breast tissue was predominantly located in the axilla, with a frequency of 71% according to Evans and Guyton [1] and 58% according to Marshall et al. [5]. The frequencies for other sites were 18.5% in the parasternal region, 8.6% in the subclavicular region, 8.6% in the submammary region, and 4% in the vulvar region [5]. Nihon-Yanagi et al. [6] reviewed 94 cases in Japan between 1978 and 2009 and reported that 91.5% of ectopic breast cancers occurred in the axilla.

Pathological findings showed similar results to anatomic breast cancers, which are mainly invasive ductal carcinomas [6,7]. FNA was used as a diagnostic tool, because it is minimally invasive. A ductal carcinoma of ectopic breast tissue is not always initially diagnosed by FNA. It is particularly difficult to differentiate ectopic breast cancer from lymph node metastasis in patients with adenocarcinoma [6]. A differential diagnosis with lymph node metastasis can be established by confirming the absence of lymph node tissue and lymphovascular infiltration. Normal breast tissue surrounding the tumor is a common pathological finding in the majority of case reports, supporting the histological conclusion that the tumor originated from ectopic breast tissue [6-8]. To this end, a core needle biopsy was performed to obtain sufficient tissue. However, in our case, normal breast tissue was not found near the tumor, conflicting with our diagnostic confirmation of the tumor originating from ectopic breast tissue. Owing to the poor histological tissue grade, the histological distinction was difficult. The differential diagnosis of a tumor originating from the skin adnexal gland was necessary. However, the tumor displayed no continuity with respect to the skin, originated from the deep layer, and proliferated throughout the subcutaneous layer. Finally, despite the absence of normal breast tissue, we concluded that the tumor had originated from the ectopic breast tissue.

Some authors recommend ipsilateral radical mastectomy as the surgical treatment of choice. However, Cogswell and Czerny [9] argued that there is no difference in the recurrence rate between radical mastectomy and local excision with axillary dissection. Mastectomy should be conducted when a differential diagnosis is difficult [10]. According to case reports from Japan from 1998 until today, local excision was conducted in 79.2% of cases, whereas mastectomy was conducted in 20.8% of cases [7]. In the present case, wide excision with a clear cut margin resulted in local recurrence and distant metastases. However, considering the rarity of the disease itself and the lack of follow-up or staging data, it was very difficult to evaluate the outcome and prognosis of either surgical treatment.

Lymph node metastasis is considered to be one of the most significant prognostic factors and is likely to occur through the surrounding anatomical structures that are in close proximity [7]. As was shown in our case, axillary node metastasis revealed a poor prognosis with rapid recurrence and distant metastases.

Initially, an axillary invasive ductal carcinoma that originates from ectopic breast tissue is prone to be misdiagnosed as a benign inflammatory disease. Delayed diagnosis is an important concern. To confirm the diagnosis, we should perform a careful evaluation using imaging and pathological testing. Then, an appropriate choice of surgical treatment is important. If advanced node metastases is present, as was the case for our patient, aggressive surgical treatment options such as a mastectomy should be performed. Due to its rarity, further studies are needed to accurately understand this disease and assess treatment results.

**CONFLICT OF INTEREST**

The authors declare that they have no competing interests.

**REFERENCES**