Breast Sarcoidosis Appearing as a Primary Manifestation of Sarcoidosis: A Case Report

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Breast involvement as the primary manifestation of sarcoidosis is rare (1, 2) and it usually shows similar radiological findings as for a breast carcinoma. The diagnosis is very difficult and tissue confirmation is necessary. Although a core needle biopsy may be helpful for diagnosis, an excisional biopsy may be needed occasionally (3, 4).

We report here on a rare case of a breast sarcoidosis that presented as a primary manifestation of sarcoidosis. Ultrasound (US)-guided directional vacuum-assisted biopsy (DVAB) was helpful for a correct diagnosis.

Case Report

A 60-year-old woman was admitted to our hospital for a palpable breast mass and newly developed dyspnea. The patient had a personal history of breast excision for a mass that was pathologically confirmed as a benign lesion several years ago. Two years ago, the patient complained of a new palpable breast mass and underwent US-guided core needle biopsy for a suspected breast malignancy. The pathological result showed chronic inflammation. After the biopsy, the patient still felt a lump and underwent repeated US-guided core biopsy for two more times. The pathological results for the mass again showed the presence of chronic granulomatous inflammation. Although acid fast staining and a polymerase chain reaction test for tuberculosis were negative, given the pathological result, the patient was presumed to be diagnosed with tuberculosis. Anti-tuberculosis medication was administered for 3 months. Despite the treatment, there was no change in the palpable breast lesion. Furthermore, the patient then complained of newly developed dyspnea and chest pain and was referred to our hospital.

The patient had neither a family nor personal history of breast cancer nor a personal history of pulmonary disease. Initial chest plain radiography (Fig. 1A) revealed numerous tiny pulmonary nodules in the both lungs with left pleural effusion. Computed tomography of the chest (Fig. 1B) showed multiple enlarged lymph nodes at the hilum, mediastinum, left infraclavicular fossa and left axillary fossa and miliary nodules diffusely scattered on both lungs. Diffuse interlobular septal thickening
with subpleural infiltration and left pleural effusion were also noted. These findings suggested the presence of hematolymphangitic lung metastasis or lymphoma or sarcoidosis. Mammography (Fig. 1C–E) showed a spiculated mass in the left upper inner quadrant, approximately 1 cm in diameter and an enlarged lymph node in the left axilla. Ultrasound (Fig. 1F) also showed a hypoechoic mass with a spiculated margin in the left upper inner quadrant and an enlarged lymph node in the left axilla (Fig. 1G). Based on these findings, the mass was thought to be a suspicious malignant breast mass (BI-RAD final assessment category 4c). Positron emission...
Fig. 1. F and G. A transverse view of sonography (1F) demonstrates a hypoechoic mass with a spiculated margin (white arrow) in the upper inner quadrant of the left breast. A previous excisional scar (open arrow) is noted at the upper portion of the mass. An enlarged lymph node in the left axillary fossa (arrowheads) (1G) is also noted.

H. A positron emission tomography scan shows fluorine-18-fluorodeoxyglucose (18-F FDG) uptake in the left breast mass (white arrow) and left axillary lymph node (asterisk). Mediastinal and hilar lymph nodes (open arrows) also show hot uptake.

I. Histopathological specimens show multiple non-necrotizing granulomas, suggestive of sarcoidosis (H & E stain × 100). A well-circumscribed non-necrotizing granuloma, consisting of epithelioid histocytes, with few lymphocytes (naked granuloma) is noted.

J. Follow-up chest radiography after steroid treatment reveals the disappearance of the previously noted numerous tiny nodular opacities and left pleural effusion.
tomography (Fig. 1H) showed hot uptake of fluorine-18-fluorodeoxyglucose (18-F FDG) in the left breast mass, a left axillary lymph node and multiple mediastinal and hilar lymph nodes.

Based on the imaging findings, the patient was presumed to have a diagnosis of breast malignancy with multiple metastases. However, the previous pathological findings did not show the presence of any malignant cells. It was decided to retrieve more large tissue for an accurate diagnosis and US-guided DVAB of the breast mass was performed instead of a 14-G core needle biopsy. A transbronchial bronchoscopic lung biopsy (TBLB) was also performed.

All of the pathological findings indicated the presence of non-necrotizing granulomatous inflammation within the breast (Fig. 1I) and lung tissue, which was consistent with sarcoidosis. A skin test for tuberculosis was negative and special stains for fungi and acid-fast bacilli were negative. Corticosteroid was used for treatment, there was rapid improvement of the symptoms and a reduction in the inflammation as noted in chest films (Fig. 1J).

Discussion

Sarcoidosis is a systemic disorder of unknown cause that is characterized by the presence of noncaseating granulomas with proliferation of epithelioid cells. Sarcoidosis commonly affects young and middle aged patients, with a slightly higher prevalence in women (5). It can involve, in order of frequency, the lungs, lymph nodes, spleen, liver, skin, eyes, muscles, bones, CNS and salivary glands (6). Involvement of the breast parenchyma is a rare manifestation of sarcoidosis, representing only approximately 1% of all cases (1). Patients with breast disease and sarcoidosis has been classified by Lower et al. (1) as sarcoidosis patients with breast cancer, breast cancer patients showing sarcoidosis-like breast reactions and sarcoidosis patients with breast granulomas. This last group was defined as breast sarcoidosis patients. Ojeda et al. (2) reviewed the medical literature of 35 cases with breast involvement of sarcoidosis from 1921 through 1997. It was reported that the presence of breast granulomas was the initial presentation in 9 out of 35 cases (2).

In a review of the literature, imaging findings of breast sarcoidosis has only infrequently been reported. Storm et al. demonstrated a case of breast sarcoidosis that presented as a well-circumscribed mass on mammography and sonography (7). The lesions have been frequently described as having imaging findings with malignant characteristics, including spiculated margins in other studies (4, 8, 9). In this case, breast sarcoidosis also showed a spiculated margin on mammography and sonography, suggestive of a breast malignancy. As breast sarcoidosis as a primary manifestation is very rare and no radiological findings can distinguish it from a breast carcinoma, a proper diagnosis is impossible to determine based on imaging findings. However, it should be considered in the differential diagnosis of spiculated breast masses, particularly in patients with systemic sarcoidosis or a history of this disease. Finally, a pathological diagnosis is mandatory.

Pathological findings for sarcoidosis consist of noncaseating granulomas with epithelioid cells and large multinucleated giant cells. However, granulomas from other inflammatory processes such as tuberculosis, histoplasmosis and fungal infections, and a tumor-related sarcoid reaction need to be excluded (10). A limited number of previous reported cases used fine needle aspiration or an automated core biopsy for a diagnosis of breast sarcoma. However, Storm et al. (7) suggested that core needle biopsy is inherently limited as a result of the potential sampling error, so the use of an excisional biopsy may be more reliable. As coexistence of sarcoidosis and breast carcinoma has also been described (1), an accurate diagnosis through larger tissue sampling is important to differentiate sarcoidosis from other granulomatous diseases and to detect a carcinoma that may co-exist with sarcoidosis. Although this the patient underwent automated core needle biopsy three times, there were inadequate pathological findings such as an indication of chronic granulomatous inflammation that resulted in the mistaken identification of the mass for tuberculosis. In Korea, when a pathological specimen shows a chronic granulomatous inflammation, it is usually presumed to be a diagnosis for tuberculosis, because tuberculosis is endemic in Korea. However, in such a case, breast sarcoidosis should be considered in the differential diagnosis and an effort for a definite diagnosis may be required. We performed an US-guided DVAB for the breast mass instead of core needle biopsy for a definite diagnosis and the breast mass was confirmed as sarcoidosis.

In conclusion, breast sarcoidosis is rare and a histological diagnosis is mandatory because no clinical or radiological findings can distinguish it from a breast carcinoma. Although 14-gauge automated core biopsy may be reliable, if it is inconclusive, US-guided DVAB may be
very helpful for a more precise diagnosis.

References