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Primary breast lymphoma clinically mimicking acute mastitis: a case report

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ABSTRACT

Extranodal non-Hodgkin lymphoma of the breast is a rare disease. We present a case of primary breast lymphoma with atypical clinical manifestations that looked like acute mastitis. A 46-year-old woman had noted a painful swelling in the right breast for 2 months. The mass had an inflammatory appearance and acute mastitis was the clinical impression. She underwent a core biopsy of the mass, and pathology showed inflammatory changes. The inflammatory mass regressed and recurred during hospitalization, and further incision with debridement was done. The histological findings were consistent with diffuse large B cell lymphoma. Systemic examination found stage IIE disease. She received chemotherapy and local radiation is planned.

Introduction

Primary breast lymphoma (PBL) is rare and accounts for 0.04-0.5% of breast malignancies and 1.7-2.2% of extranodal non-Hodgkin lymphomas[1,2]. The diagnostic criteria for PBL are 1) presence of technically adequate pathological specimens, 2) close association of mammary tissue and lymphomatous infiltrate, 3) no prior diagnosis of an extrammamary lymphoma, and 4) no evidence of concurrent widespread disease, except for ipsilateral axillary lymph nodes if concomitant with the primary lesion[3]. The majority of cases are B-cell lymphomas and the most common histological type is diffuse large B-cell lymphoma[4,5]. We would like to present an interesting case of PBL with uncommon clinical manifestations. In most reports the clinical presentation was a mass lesion, but our patient had diffuse inflammation mimicking severe mastitis.

Case presentation

A 46-year-old woman visited our hospital complaining of severe epigastric pain and vomiting for one day and the presence of an inflammatory swelling in her right breast for 2 months. Physical examination found erythematous changes and induration on the right breast with tenderness. The abdomen showed diffuse tenderness and rebound pain with hypoactive bowel sounds. There was no fever, and laboratory tests gave a white blood cell count of 14,470/mL with 72.8% neutrophils; the C-reactive protein value was 15 mg/dL. Post-contrast computed tomography revealed severe bowel wall thickness in the jejunum, and acute mesenteric ischemia was strongly suspected.

The patient underwent an exploratory laparotomy with resection of a gangrenous small bowel about 150 cm in length. A core biopsy of the right breast mass was done. Macroscopically, the breast had an inflammatory appearance. Pathological examination of the small bowel revieled acute and chronic inflammation and ischemic enteritis. The right breast mass also showed acute and chronic inflammato-

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ry changes histologically. During hospitalization, the inflammatory mass of the right breast regressed. However, it recurred like the initial presentation a couple of days later. The patient underwent an incision and drainage with debridement of the right breast. The impression was still of acute mastitis. A computed tomography scan also showed an inflammatory-like mass in the right breast (Figure 1). Pathology revealed diffuse infiltration of small and large atypical lymphocytes with irregular nuclei (Figure 2A). Immunohistochemistry was negative for cytokeratin, CD56 and CD3, and positive for CD20 (Figure 2B). The histological findings were consistent with a diagnosis of diffuse large B-cell lymphoma.

Ultrasonography demonstrated a large heterogeneously hypodense lesion in the right breast. Enlarged right axillary lymph nodes were also detected. The impression was a that of a breast tumor with right axillary lymphadenopathy. The LDH level was 461 IU/L and a gallium scan showed that the lesions were confined to the right breast and axillary region. Bone marrow biopsy was negative. The patient did not have B symptoms. The final diagnosis was primary right breast lymphoma, diffuse large B-cell type, stage IIE. She has just finished 4 cycles of chemotherapy with cyclophosphamide, daunorubicin, vincristine and prednisolone, which was associated with grade 4 acute hematological toxicity (hemoglobin 5.6 g/dL; NCI Common Toxicity Criteria version 2.0). A nearly complete response was achieved. The patient will receive local radiotherapy to the right breast and regional lymph nodes. The prescribed radiation dose is 46.8 Gy in 26 fractions. Three-dimensional conformal radiotherapy with bilateral opposed tangential photon fields and an anterior-posterior field will be used to cover her right breast plus the axillary and supraclavicular lymph nodes.

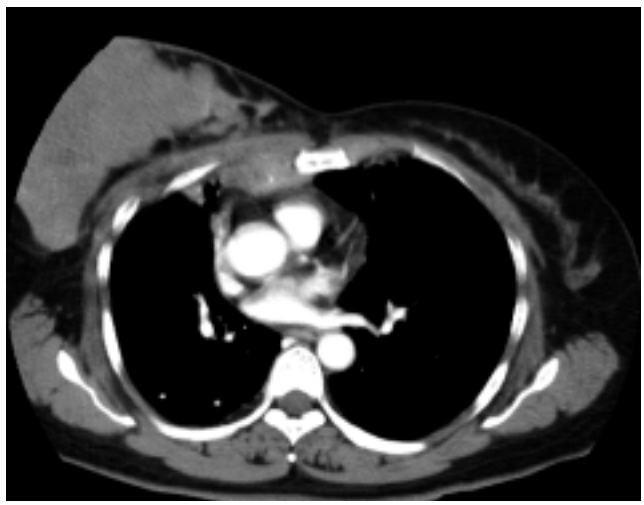

Figure 1 - Computed tomography showing a breast mass.

Discussion

The breast is a rare primary site for extranodal malignant lymphoma. Wiseman and Liao[3] defined PBL pathologically as the close association of mammary tissue and lymphomatous infiltrate with neither a prior diagnosis of extrammamary lymphoma nor evidence of concurrent widespread disease except for ipsilateral axillary lymph nodes[3]. Our patient's lesion fit the criteria for PBL, and there were no extramammary sites of involvement other than the ipsilateral lymph nodes.

PBL does not have typical clinical features and cannot be predicted preoperatively based only on clinical and radiological findings. Breast carcinoma, fibroadenoma, and phyllodes tumor might be the preoperative diagnoses[5,6]. Grubstein *et al.*[7] reported on a 22-year woman with extranodal primary B-cell non-Hodgkin lymphoma of the right breast mimicking acute mastitis. The clinical pattern looked like acute inflammatory changes, and the sonographic presentation of their case was not of a mass

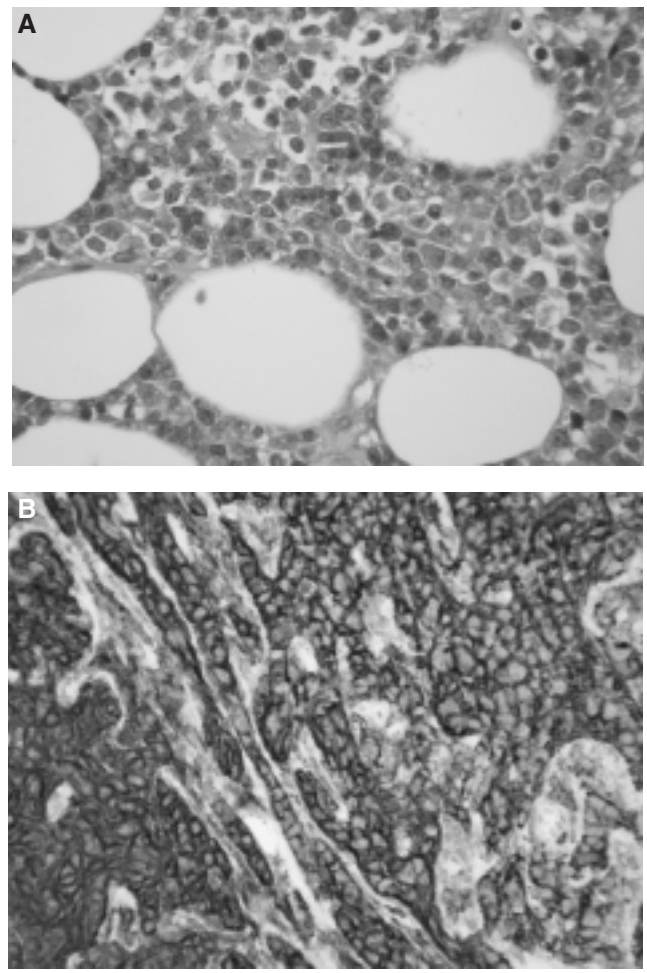

Figure 2 - A) Pathology of the right breast mass revealed diffuse large B lymphocytes. B) Immunohistochemistry showed positivity for CD20.

but an infiltrating anechoic process mimicking mastitis, although computed tomography showed the solid nature of the lesion. Typical sonographic findings for PBL are single, circumscribed or microlobulated and oval masses, and the echo pattern of the mass is usually hypoechoic[8,9]. Our patient showed an acute mastitis pattern in her right breast, which was compatible with the imaging results. The final diagnosis, however, was based on the pathology findings. As stated by Grubstein *et al.*[7], "lymphoma is often referred to as the great imitator; whenever there is a mismatch between the physical findings and imaging findings, lymphoma should be considered in the differential diagnosis."

Jeanneret-Sozzi *et al.*[10] conducted a study on a large series of PBL cases. They collected 84 consecutive patients who were treated in 20 institutions of the Rare Cancer Network. The most common symptom was a palpable mass (61%), followed by palpable lymph nodes (25%). However, there were still 9 patients with local signs of inflammation. Prior to this study, another study with an even larger sample size had been performed. Ryan *et al.*[11] collected 204 eligible patients presenting to the International Extranodal Lymphoma Study Group (IELSG)-affiliated institutions from 1980 to 2003. None were reported to have an inflammatory presentation. Both studies showed similar patient characteristics. The median age of the patients was 64 years. More than 94% of the cases were stage IE or IIE, and pathology was diffuse large B-cell lymphoma in the majority. Our patient was younger (46 years), but the stage and pathology are consistent with the findings of the above review.

Appropriate treatment guidelines have not been well identified because of the limited data available. However, we can glean some consensus from the literature. Mastectomy does not appear to have any benefit, and surgery should be limited to biopsy to establish the correct histological diagnosis. The main treatment modality is anthracycline-containing chemotherapy, especially for intermediate- and high-grade histology. Local irradiation can increase the local control rate and probably the survival rate[10-14]. The treatment of our patient is in accordance with the above suggestions.

The risk of central nervous system (CNS) relapse varies between studies. Some authors emphasized that the CNS was a major site of relapse in PBL[1,10,12,14,15]. They observed a high incidence of CNS relapse in this group of localized extranodal lymphoma. However, the largest retrospective study by the IELSG did not show the CNS as a major site of relapse[11]. The role of CNS prophylaxis remains uncertain, and this is why we did not plan prophylactic radiotherapy to the CNS in our patient.

In conclusion, PBL is relatively uncommon and has no typical clinical or imaging features. It may physically manifest like acute mastitis. Lymphoma should be taken into account as a possible diagnosis in cases that do not settle rapidly with appropriate antibiotics, and histological examination is essential in such cases. The optimal treatment is still undetermined, but the usefulness of combined-modality treatment (chemotherapy and local radiation) is suggested. The benefit of CNS prophylaxis is yet to be proven.

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