

Gallium-67 scintigraphic findings in a patient with breast lymphoma complicated with Sjögren syndrome

Mana YOSHIMURA,* Kiyoshi KOIZUMI,* Kenichiro SATANI,* Dai KAKIZAKI,*
Yoshikazu KAWANISHI,** Kazuma OHYASHIKI** and Kimihiko ABE*

*Departments of *Radiology and **Internal Medicine, Tokyo Medical University*

We report here a patient with mucosa associated lymphoid tissue (MALT)-lymphoma of the breast complicated with Sjögren syndrome. It is speculated that Ga-67 could accumulate not only in lymphoma lesions but also in benign lymphoproliferative locations of Sjögren syndrome. Gallium-67 scintigraphy might be useful for the diagnosis and therapeutic monitoring of MALT-lymphoma complicated with Sjögren syndrome.

Key words: gallium-67, breast lymphoma, mucosa associated lymphoid tissue, Sjögren's syndrome

INTRODUCTION

MALIGNANT LYMPHOMA of the breast is rare since the incidence is 0.04% to 0.53% of all malignant neoplasms of the breast and 2% of extranodal malignant lymphoma.¹ Sjögren syndrome (SJS) is an autoimmune disease, and occurs primarily in the salivary and lacrimal glands, though almost half of the patients progressively develop generalized autoimmune disease with extranodal expansion of T and B cells. Malignant lymphoma occurs in SJS with a frequency of 5% based on benign lymphoepithelial lesion (LEL) or myoepithelial sialoadenitis (MESA).² This type of malignant lymphoma is considered to be a kind of mucosa associated lymphoid tissue (MALT)-lymphoma. As far as we know there is no case report of breast lymphoma complicated with SJS in the literature. We present the first case report with special reference to its diagnosis and therapeutic monitoring by gallium-67 scintigraphy.

CASE REPORT

A 30-year-old female was admitted to our hospital after

detailed examination a mass in the right breast on October 1998, because lymphohyperplasia had been detected by fine-needle aspiration cytology of the mass. Two months prior to admission, she presented with swelling of the uvula, and uvulectomy revealed MALT-lymphoma (B cell type diffuse small cell lymphoma) of the minor salivary gland. In addition, 4 months earlier she had delivered a child prematurely. On admission, eruptions were noticed all over her body and were considered to be the atopic dermatitis. Physical examination revealed a mass, of 4 cm inside diameter, in the right breast and enlargement of both parotid glands without pain. The remainder of the physical examination was unremarkable. She was again diagnosed as having MALT-lymphoma by open biopsy of the breast mass. Chest enhanced CT demonstrated a well circumscribed mass lesion in the central and medial area of the right breast (Fig. 1). CT scan of the neck showed enlargement and heterogeneous parenchymal enhancement with multiple low attenuating lesions of both parotid glands indicating sialoadenitis and/or a lymphoproliferative disorder possibly due to SJS (Fig. 2). First gallium-67 scintigraphy for staging was done on November 1998. Significant tracer accumulation was demonstrated in the central and medial area of the right breast corresponding to the mass detected by CT, as well as a horseshoe-like accumulation in both breasts. There was no abnormality elsewhere except in the left parotid gland (Fig. 3a). The clinical stage was determined to be IIa according to the Ann-Arbor classification.

Received February 17, 2000, revision accepted April 10, 2000.

For reprint contact: Mana Yoshimura, M.D., Department of Radiology, Tokyo Medical University, 6-7-1 Nishi-shinjuku, Shinjuku-ku, Tokyo 160-0023, JAPAN.

Although she did not have any subjective siccant complaints, immunological examinations and tests for siccant syndrome were performed. The results were as follows: antinuclear antibody (ANA) 1: 320 with a speckled pattern, anti-SS-A/Ro antibody 1: 256, and positive result of Schirmer's test. Therefore a diagnosis of subclinical SJS was made. Based on these findings the patient was diagnosed as having MALT-lymphoma of the breast and subclinical SJS, and was treated with CHOP (cyclophosphamide, doxorubicin, vincristine, prednisone) chemo-

therapy. After 3 courses of CHOP chemotherapy, the palpable mass lesion in the right breast and swelling of both parotid glands had disappeared on physical examina-

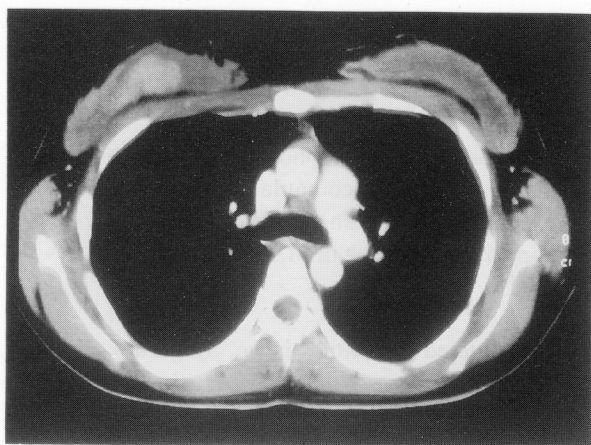


Fig. 1 CT scan of the breast (Pre-chemotherapy). Well circumscribed mass lesion is noted in the central and medial area of the right breast.



Fig. 2 CT scan of the neck (Pre-chemotherapy). Enlargement and heterogeneous parenchymal enhancement with multiple low attenuating lesions of the bilateral parotid glands represents sialoadenitis and/or lymphoproliferative disorder due to SJS.

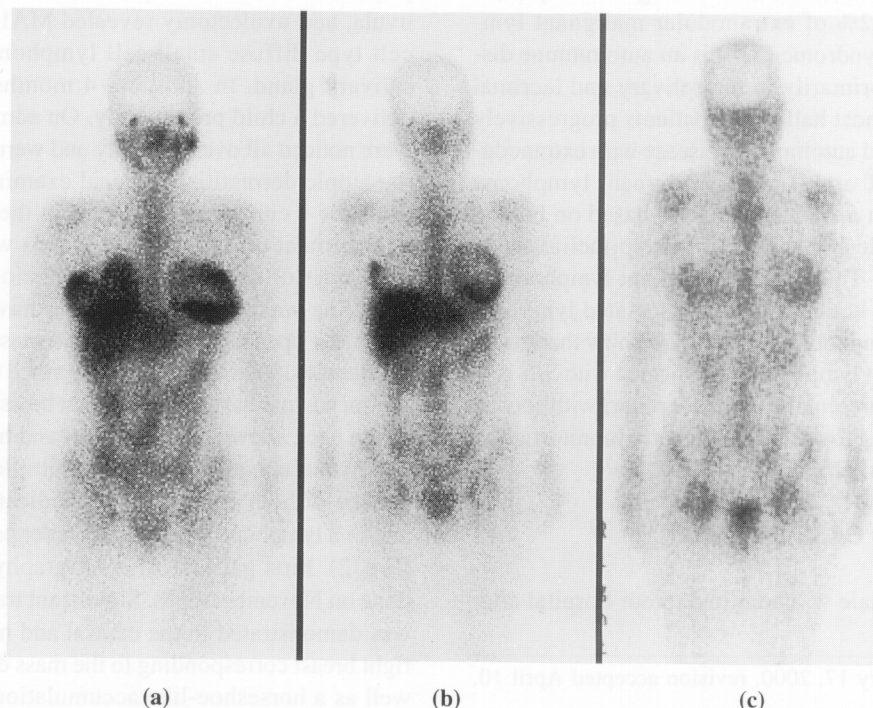


Fig. 3 Gallium-67 scintigraphy (a) Pre-chemotherapy. Significant tracer accumulation is shown in the bilateral breast and left parotid gland. (b) (c) After 3 and 5 courses of chemotherapy. Accumulation of the tracer reduced gradually.

tion and follow-up CT examination. The Ga-67 accumulation in the breast had also disappeared on the mass and decreased in the horseshoe-like area in January 1999 (Fig. 3b). The findings of immunological examinations were also reduced (antinuclear antibody 1: 40, anti-SS-A/Ro antibody 1: 64). The next 2 CHOP courses were then added. The third gallium-67 scintigraphy was done on March 1999, and the accumulation in the horseshoe-like area was also reduced (Fig. 3c). The patient was monitored for 15 months altogether, and maintained complete remission.

DISCUSSION

Breast lymphoma is clinically and histologically classified into two groups. One is Burkitt's type lymphoma, which occurs bilaterally in the young. The other is B-cell lymphoma that occurs in the elderly. Although the latter is now considered to be one kind of MALT-lymphoma, its pathological classification and treatment is still controversial.^{3,4}

In this patient, because MALT-lymphoma was revealed in the uvula and breast, it is concluded that multicentric MALT-lymphoma originated in salivary gland and breast based on the generalized autoimmune disorder complicated with subclinical SJS.

Gallium-67 scintigraphy is known to accumulate in various tumors, including lymphoma. In this patient, this examination was performed 3 times, pre-chemotherapy and after 3 and 5 courses of chemotherapy. In the first scintigraphy, an abnormal uptake to the right breast tumor in the central and medial area was pointed out. Subsequent scintigraphic finding revealed reduced accumulation in the lesion after 3 courses of chemotherapy, when no palpable mass lesion was detected by physical examination or CT, confirming the previous finding as abnormal uptake to the tumor. The horseshoe-like accumulation in both breasts seen on the first scintigram became less intense on the second scintigram and even less on the third one. There are three hypotheses concerning this distribution of Ga-67. First, diffuse infiltration was on bilateral

breasts in addition to the palpable mass lesion. Second, accumulation to a lymphoproliferative disorder such as benign LEL or prelymphomatous lesion on bilateral breasts was reduced gradually by chemotherapy. This view is supported by the diminishing of the sialoadenitis and/or lymphoproliferative disorder due to SJS in the left parotid gland after chemotherapy. It is reported that Ga-67 accumulates in lymphoproliferative lesions due to SJS.⁵ Third, nonpathologic accumulation in the breast postpartum was reduced naturally or for some other reason. Increased uptake to the postpartum breast often has a doughnut-like shape bilaterally, and is considered to continue for a few months.⁶ Although the third hypothesis is the most likely to account for the horseshoe-like accumulation in both breasts, the exact mechanism of Ga-67 uptake to the breast outside the mass is not known because histological confirmation was not done except for the mass.

In summary, we report a patient with MALT-lymphoma of the breasts complicated with SJS. It is speculated that Ga-67 might accumulate not only in the lymphoma but also in a benign lymphoproliferative lesion due to SJS.

REFERENCES

1. Kanda Y. Primary breast lymphoma. *Hematology & Oncology* 39: 137-142, 1999.
2. Sugai S. Salivary gland. *Hematology & Oncology* 39: 115-124, 1999.
3. Lamovec J, Jancar J. Primary malignant lymphoma of the breast. Lymphoma of the mucosa-associated lymphoid tissue. *Cancer* 60: 3033, 1987.
4. Aozasa K, Ohsawa M, Saeki K, Horiuchi K, Kawano K, Taguchi T. Malignant lymphoma of the breast. Immunologic type and association with lymphocytic mastopathy. *Am J Clin Pathol* 97: 699, 1992.
5. Collins RD, Ball GV, Logic JR. Gallium-67 scanning in Sjögren's syndrome: Concise communication. *J Nucl Med* 25: 299-302, 1984.
6. Kim YC, Brown ML, Thrall JH. Scintigraphic patterns of Gallium-67 uptake in the breast. *Radiology* 124: 169-175, 1977.