Case Report

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Secondary Breast Burkitt Lymphoma Involving the Stomach, Ovary, Pancreas, and Bones: A Case Report

전신의 다발성 침범을 동반한 이차성 유방 버킷 림프종: 증례 보고

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Breast lymphomas are rare, malignant breast neoplasms with a heterogeneous pattern of clinical symptoms. Burkitt's lymphoma is a rare, highly aggressive, and rapidly growing B-cell non-Hodgkin lymphoma. We report about a 27-year-old woman diagnosed as having secondary breast Burkitt's lymphoma, probably originating from the stomach, with multiple distant metastases. Breast ultrasonography revealed multiple, variable sized, heterogeneous masses with posterior acoustic enhancement and echogenic rims. These imaging findings may sometimes overlap with those of other breast malignancies. However, unlike other breast malignancies, lymphoma can be diagnosed by biopsy and does not require surgical excision. To avoid unnecessary treatment, radiologists and clinicians should be aware of the characteristic imaging features of breast lymphomas.

Index terms Breast; Burkitt Lymphoma; Diagnostic Imaging; Lymphoma; Ultrasonography

INTRODUCTION

Breast lymphomas are very rare and account for only 0.04%–0.7% of all malignant breast neoplasms (1, 2). Breast lymphomas can be classified as primary or secondary. Secondary lymphomas are the most common metastases to the breast, accounting for 17% of all breast metastases (1, 3), and are more common than primary

lymphomas (4). They are difficult to diagnose because of nonspecific imaging findings and overlapping imaging features with other breast malignancies. Burkitt lymphoma is a rare, highly aggressive, and rapidly growing B-cell non-Hodgkin lymphoma (NHL). Here, we report about a 27-year-old woman diagnosed as having secondary breast Burkitt lymphoma, probably originating from the stomach, with multiple distant metastases.

CASE REPORT

A 27-year-old woman complaining of abdominal distension that had persisted for 1 week presented to our hospital. The patient had no other medical problems. As an initial imaging procedure, abdominopelvic CT revealed huge bilateral masses in both ovaries with ascites. Subsequently, ^{18}F fluorodeoxyglucose PET-CT revealed multifocal hypermetabolic lesions in the stomach, ovaries, breasts, pancreas, thoracic vertebrae, sacrum, left ilium, and both femurs (Fig. 1A). Endoscopy revealed multiple ulcerofungating masses in the stomach, and a biopsy was performed. Pathological immunostaining results showed positive reactions for c-Myc, CD20, and Bcl6, confirming Burkitt's lymphoma. Based on the patient's initial complaint of abdominal distension and since primary Burkitt lymphomas most commonly involve the gastrointestinal (GI) tract, our presumptive diagnosis was primary gastric Burkitt lymphoma (5).

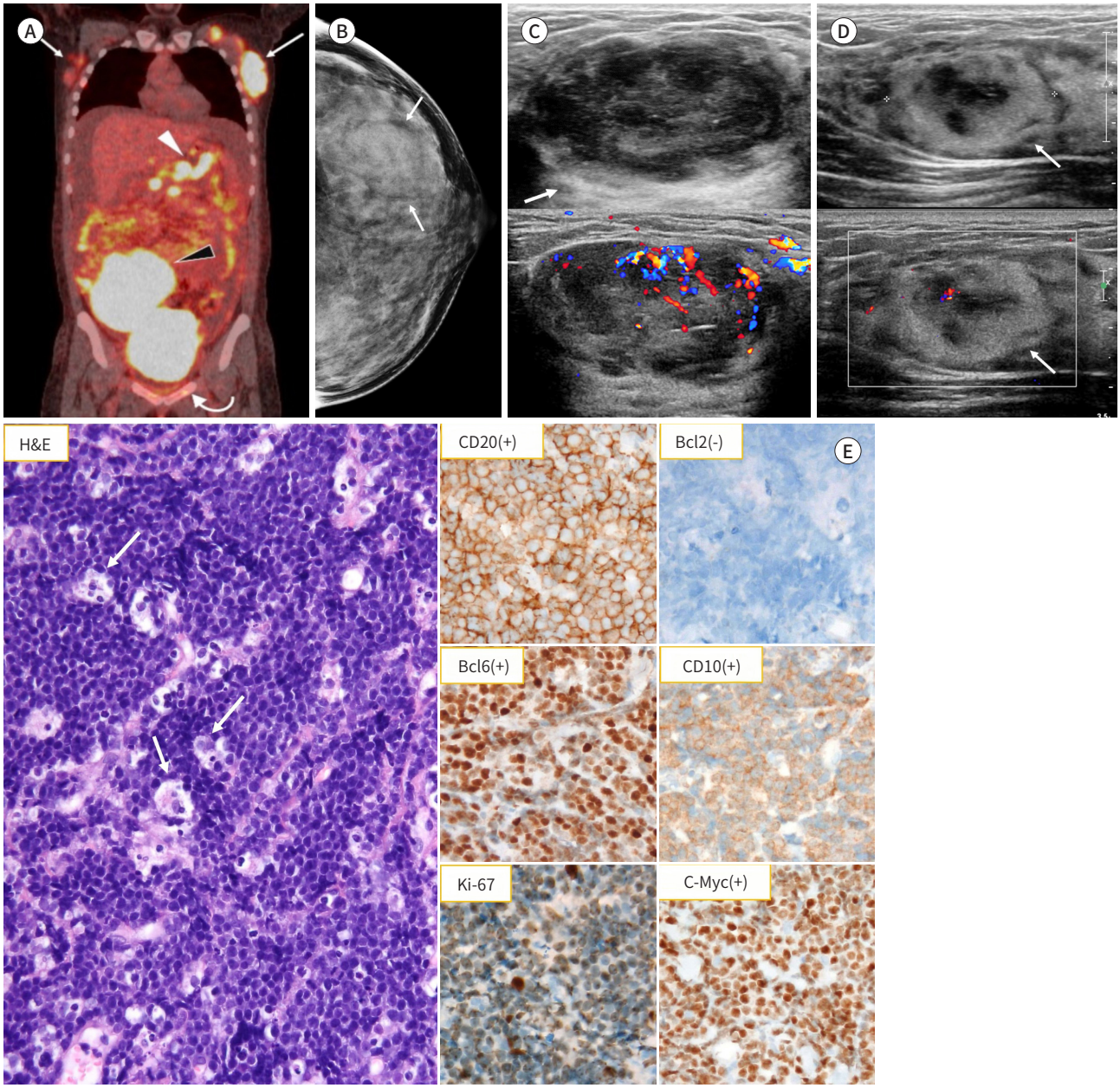
Physical examination revealed a palpable mass in the left breast. The patient had a family history of maternal breast cancer. The patient underwent mammography and breast US for differential diagnosis.

On mammography, an oval, obscured, isodense mass was suspected in the left upper outer area; however, the evaluation of this lesion was limited because of extremely dense breast tissue (Fig. 1B). Therefore, breast US was recommended for an accurate evaluation.

Breast US revealed multiple, variously sized, heterogeneous, hypoechoic masses in both breasts. The largest mass (5 cm in diameter) was in the left upper outer breast (at 2 o'clock position) and appeared oval, indistinct, and hypoechoic, with increased vascularity and posterior acoustic enhancement (Fig. 1C). Another mass that was oval, indistinct, and heterogeneous with an echogenic rim was noted (Fig. 1D). US-guided biopsy was performed on the largest breast mass. Hematoxylin and eosin staining of the specimen (× 200) showed small monotonous cells with a high nuclear/cytoplasmic ratio and a characteristic "starry sky pattern" due to the presence of scattered histiocytes engulfing the apoptotic lymphoma. Immunohistochemical CD20 marker staining (× 200) was positive, and a specific finding for B-cell lymphoma. Negative Bcl2 marker staining (× 200) and positive Bcl6 (× 200) can rule out diffuse large B-cell lymphoma, which is the most common of B-cell lymphomas. c-Myc positivity (× 200) is a characteristic marker for Burkitt lymphoma (Fig. 1E). Therefore, the patient was diagnosed with secondary breast lymphoma. Although the patient was scheduled for the chemotherapy, follow-up information was not available.

The need to obtain written informed consent was waived by the Institutional Review Board due to the retrospective nature of this report (IRB No. 2023-05-024).

Fig. 1. A 27-year-old woman having secondary breast Burkit lymphoma presenting with abdominal distension and palpable lump in the left breast.
A. ^{18}F-FDG PET/CT shows multiple hypermetabolic lesions in the stomach (white arrowhead), both ovaries (black arrowhead), both breasts (arrows), pancreas, thoracic vertebra, both sacrum, left ilium (curved arrow) and both femurs.
B. Mammography shows oval, obscured, isodense masses (arrows) in the upper, outer quadrant of the left breast.
C. US reveals a 5-cm-sized oval indistinct heterogeneous, hypoechoic mass with increased internal vascularity and posterior acoustic enhancement (arrow) in the left, upper, outer breast (2 o'clock position).
D. Another mass reveals as an oval, indistinct, heterogeneous mass with an echogenic rim (arrows) in the left inner breast.
E. Histologic specimen shows small monotonous cells with high nuclear/cytoplasmic ratio and characteristic "starry sky pattern" due to the presence of scattered histiocytes (arrows) engulfing apoptotic lymphoma (H&E, × 200). Immunohistochemical CD20 marker staining was positive, and is a specific finding for B-cell lymphoma. Results of negative Bcl2 marker (× 200) and positive Bcl6 (× 200) ruled out diffuse large B-cell lymphoma, which is the most common of B-cell lymphomas. CD10 marker positivity and high proliferation index on Ki-67 marker staining are also shown. C-Myc positivity is a characteristic marker for Burkitt lymphoma.
H&E = hematoxylin and eosin



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DISCUSSION

Burkitt lymphoma is a rare and highly aggressive form of B-cell NHL. It is characterized by the translocation and deregulation of the *MYC* gene on chromosome 8 and has the potential to involve multiple organ systems (5). Burkitt lymphoma has three subtypes (sporadic, endemic, and immunodeficiency-associated), each demonstrating different epidemiology, risk factors, and clinical presentations (5).

In this case, the sporadic subtype was considered. This subtype has an overall incidence of three cases per million per year in the general population. This type of NHL is relatively more common in the pediatric population, accounting for 30% of childhood lymphomas and less than 1% of adult NHLs (5). Sporadic cases have been associated with the Epstein-Barr virus, which was negative in our patient. The abdomen, particularly the GI tract, is the most common site of Burkitt lymphoma development (5). Our patient showed involvement not only in the stomach but also in the gonads and breasts, which is relatively uncommon (5).

Breast Burkitt lymphoma mostly affects young women of childbearing age (range 12–36 years). Moreover, cases often present during pregnancy or lactation in the postpartum period (6, 7). Patients may complain of a rapidly growing, unilateral or bilateral, well-circumscribed breast mass as their first symptom (7).

On mammography, breast lymphoma usually presents as a solitary or multiple, non-calcified, circumscribed, oval or round mass of variable density, and may also be observed in the form of ill-defined diffuse attenuation (2, 8). Unfortunately, mammography has a limited ability to detect lymphomas that appear as diffuse infiltrates or small masses. Therefore, US may be a more suitable modality for detecting these lesions.

Breast lymphoma usually appears as a hypoechoic solid mass with circumscribed or indistinct margins. Mixed internal echogenicity and hyperechogenicity are also observed frequently in breast lymphoma (1, 2). The hyperechoic nature of breast lymphoma likely reflects tumor cells intermingled with adipose tissue. The central area consisted of tumor cells within dense stroma and the hyperechoic rim correlated histologically with tumor cells infiltrating into adipose tissue at the periphery (9). Posterior acoustic enhancement and an echogenic rim, or onion-pill like rim, surrounding the lesion are common features that represent lymphedema (2). When composed of homogeneous cells organized in a round structure, lymphoma can produce posterior acoustic enhancement (10). These findings were also observed in our patient and may sometimes overlap with those of other breast malignancies, such as melanoma or metastatic lesions from other sites. The multiple bilateral distributions of the lesions support the possibility of lymphoma involvement. US is a useful modality for both diagnosis and imaging-guided biopsies. Because lymphoma does not require surgical resection, unnecessary invasive treatments should be avoided using image-guided biopsies.

PET/CT is a useful modality for evaluating the systemic involvement of lymphoma and staging. It is excellent for identifying lymph node/extranodal involvement, radiotherapy planning, and treatment response monitoring.

Chemotherapy is the primary treatment for Burkitt lymphoma. Owing to the lack of randomized controlled trials, there is no clear choice of therapy. However, current treatment protocols generally employ intensive multi-agent regimens (5).

In conclusion, breast lymphomas, including Burkitt lymphoma, may have a heterogeneous pattern of clinical symptoms and course. Breast lymphoma usually appears as a hypoechoic or heterogeneous echogenic solid mass with circumscribed to indistinct margins and multiple bilateral distributions. Posterior acoustic enhancement and an echogenic rim may be depicted. These imaging findings may sometimes overlap with those of other breast malignancies. However, unlike other breast malignancies, lymphoma can be diagnosed through biopsy and does not require surgical excision. It should be treated with chemotherapy, immunotherapy, irradiation, or stem cell transplantation. To avoid unnecessary treatment, radiologists and clinicians should be aware of this rare condition and be able to identify the characteristic imaging features of breast lymphomas.

Author Contributions

Conceptualization, L.H.J., S.G.W., P.Y.M., S.M., P.J.H.; investigation, all authors; supervision, S.G.W., P.Y.M., P.H.Y.; writing—original draft, L.H.J., P.H.Y.; and writing—review & editing, S.G.W., P.Y.M., S.M., P.J.H.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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전신의 다발성 침범을 동반한 이차성 유방 버킷 림프종: 증례 보고

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유방 림프종은 희귀한 유방 악성 종양으로 임상 증상과 경과가 이질적인 패턴을 보인다. 그 중 버킷 림프종은 매우 드물고, 공격적이며 빠르게 성장하는 B세포 비호지킨 림프종이다. 우리는 위에서 시작된 것으로 추정되고, 여러 부위의 전이를 동반한 이차성 유방 버킷 림프종으로 진단받은 27세 여성의 사례를 보고한다. 유방 초음파상에서 후방 음영 증가와 에코성 띠를 포함한 다양한 크기의 비균질한 병변으로 관찰되었다. 이러한 영상 소견은 종종 다른 유방의 악성 종양 소견과 혼동될 수 있다. 그러나 다른 유방 악성 종양과 달리 림프종은 생검을 통해 진단할 수 있으며 외과적 절제가 필요하지 않다. 불필요한 치료를 피하기 위해서 영상의학과 전문의와 임상의는 유방 림프종의 특징적인 영상학적 특징을 알아야 한다.

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