



A Case Report of Male Occult Breast Cancer Manifested as Superior Vena Cava Syndrome

상대 정맥 증후군으로 발현된 남성 잠재성 유방암의 증례 보고

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Occult breast cancer (OBC) is an asymptomatic condition. We report a case of OBC in a 72-year-old male patient who presented with severe dyspnea and upper extremity swelling. Computed tomography scan showed multiple lymph node enlargements in the supraclavicular and infraclavicular areas, and ultrasound scan showed multiple axillary lymph node enlargements. Based on the radiological findings, clinicians suspected lymphoma or disseminated metastasis from unknown primary malignancies. However, the axillary biopsy specimen revealed invasive breast cancer with hormonal receptor positivity.

Index terms

Axilla
Breast Neoplasm
Lymphatic Metastasis
Male
Occult Primary Neoplasm

INTRODUCTION

Occult breast cancer (OBC) is a rare type of breast cancer in women without any symptoms in the breast and may manifest as axillary metastasis without any malignant lesions in either breast. Male breast cancer accounts for less than 1% of all breast cancers worldwide and less than 0.6% in Korea (1). The peak age of incidence for men is 71 years, whereas for women is 52 years. The most common presentation is a palpable mass, while skin thickening and nipple retraction may also be present. Specifically, palpable axillary lymph nodes are present in about 50% of cases (2). Our case report describes a male patient diagnosed with OBC that was initially manifested as severe dyspnea accompanied by concomitant multiple lymph node enlargements in both supraclavicular and axillary areas.

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CASE REPORT

A 72-year-old man, with a 2-week history of severe dyspnea and upper extremity swelling, was referred from a local hospital to our oncology department for further examination. Physical examination showed multiple palpable masses in both axillary areas and along the neck, as well as severe edema, symptoms associated with superior vena cava syndrome. The patient was a nonsmoker and had no history of disease other than hypertension, or known family history of cancer.

Computed tomography of the chest, abdomen, pelvis, and neck revealed multiple variable-sized lymph nodes, with or without necrosis, in the supraclavicular, mediastinal, and bilateral axillary areas. In addition, diffuse subcutaneous edema was detected along the chest wall and upper extremity, in addition to multifocal enhancing lesions in both the paraspinal muscles and trapezius. Magnetic resonance imaging of breast showed

that both mammary glands were free of lesions, apart from enlarged axillary lymph nodes and subcutaneous edema. Ultrasonography revealed multiple enlarged lymph nodes with subcutaneous edema in both breasts, but no definite malignant breast mass was detected (Fig. 1). Disseminated lymphatic metastasis, lymphoma, or disseminated infectious conditions were included in the differential diagnosis on the basis of the radiologic

evaluations. An axillary lymph node was biopsied and pathological evaluation confirmed invasive ductal carcinoma. Immunohistochemistry showed the following results: estrogen receptor (ER) positivity (20%), progesterone receptor (PR) negativity, human epidermal receptor 2 (Her-2; 2+), and Ki-67 (60%). Fluorescent in situ hybridization could not confirm Her-2 gene amplification. Routine hematological and biochemical parameters

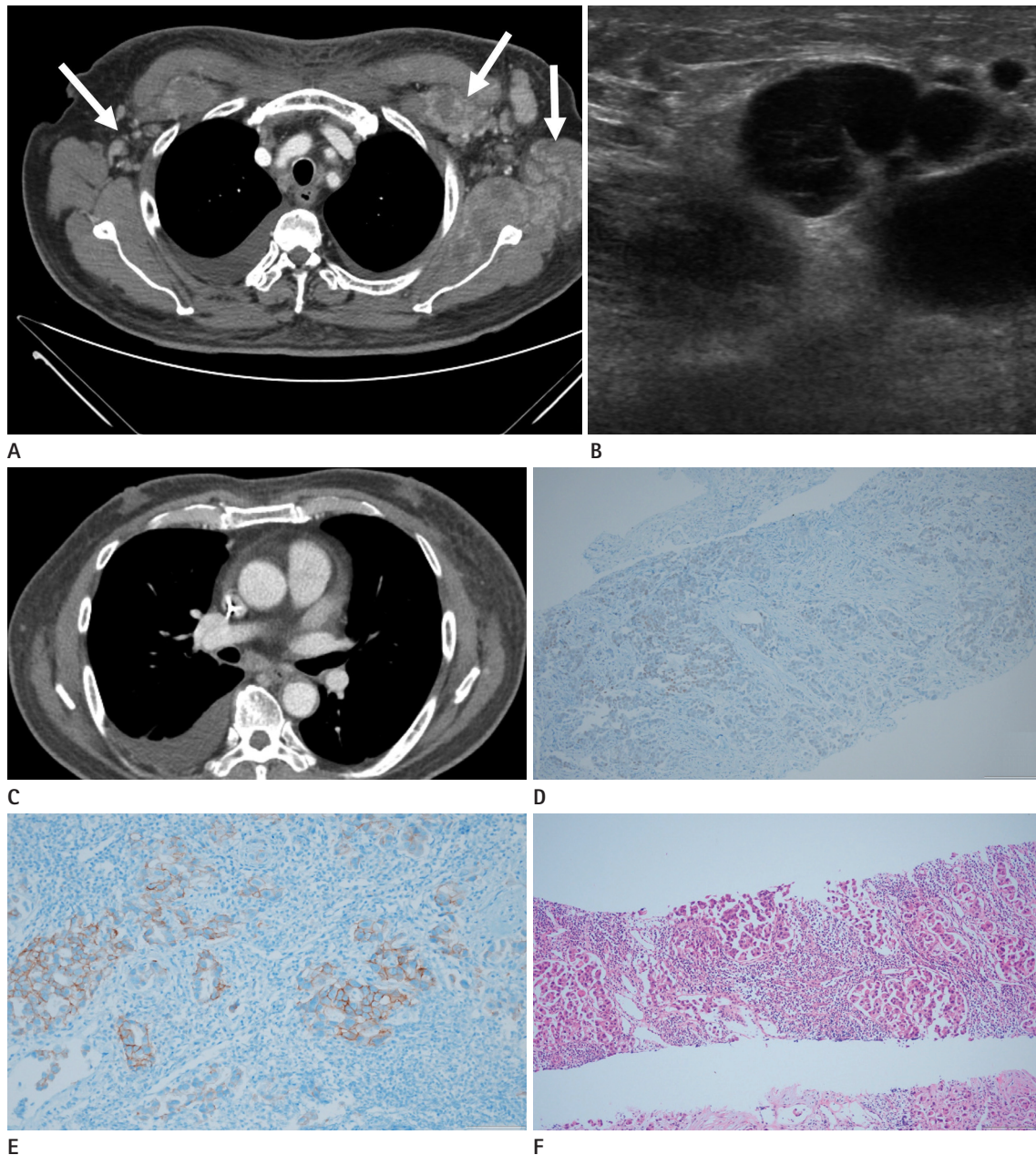


Fig. 1. A 72-year-old man who presented severe dyspnea and upper extremity swelling. CT shows **(A)** bilateral axillary lymph nodes, several enhanced intramuscular masses (arrows, **A**). Ultrasonography and CT reveal **(B)** enlarged abnormal lymph nodes in both axilla **(B)** and diffuse edematous changes **(C)**, with dilated lymphatics in both breasts. Immunohistochemical staining showing positive expression of ER, 20% ($\times 200$) **(D)** and Her-2 ($\times 400$) **(E)**, and hematoxylin and eosin ($\times 200$) staining showing metastatic lymph nodes from invasive ductal carcinoma **(F)**. CT = computed tomography, ER = Estrogen receptor, Her-2 = Human epidermal receptor 2

were within the normal range (normal range, 0–26.4 U/mL).

On the basis of all the above findings, the patient was diagnosed with OBC. The patient received neoadjuvant chemotherapy (290 mg paclitaxel and 600 mg trastuzumab every 3 weeks) without the use of any surgical treatment, such as modified radical mastectomy or axillary dissection, as he was ineligible for surgery. The patient's dyspnea resolved after treatment with chemotherapy.

DISCUSSION

In female patients with breast cancer, approximately 0.2–0.9% of cases are occult, while male breast cancer accounts for approximately 0.7% of all breast cancers worldwide (3). The rate of diagnosis of OBC manifesting as axillary lymph node metastasis is very low, with most cases being detected at a more advanced stage. Recent epidemiologic data suggest that the incidence of male breast cancer is increasing steadily (4). However, OBC in men is still rare and is often diagnosed based on symptoms of palpable lymph node metastasis in the axillary area, supraclavicular fossa, or infraclavicular fossa as the first clinical manifestation (5).

The most commonly reported palpable axillary masses are metastatic lymph nodes associated with breast cancer, followed by lung, prostate, and testicular cancer, melanoma, and squamous cell cancer. Lymphoma can also metastasize to the axillary lymph nodes. In approximately 50% of cases of OBC, the cancer origin cannot be found in the breast specimen (2). Systemic physical examination, tumor marker determination, and imaging examinations should be performed to identify the origin of the primary malignancy and make an accurate diagnosis for better patient management. In the present case, clinical and radiologic results suggested “lymphoma” or “metastasis from another primary organ” as differential diagnosis owing to the presence of multiple enlarged lymph nodes at several sites, including the supraclavicular, mediastinal, and axillary areas on both sides, as well as enhanced intramuscular masses. However, immunohistochemistry showed the presence of ER and Her-2, which is the most relevant information for the diagnosis of OBC. The presence of hormonal receptors is used not only to assess the effectiveness of adjuvant hormonal therapy but also to evaluate disease prognosis. Moreover, the presence of hor-

monal receptors also helps in the diagnosis of OBC, as in our case, when no other abnormal findings of metastasis to the axillary lymph nodes are detected. In male breast cancer, ER and PR are expressed in more than 90% and 80% cases, respectively (6). Additionally, Her-2 is useful for the differential diagnosis of breast cancer when a primary lesion cannot be confirmed. Only a few cases of male breast cancer have shown Her-2 positivity with no favorable outcomes reported when trastuzumab-containing chemotherapy was used in the neoadjuvant setting (7).

No studies have determined the optimal therapy for OBC in men, and most treatments are based on clinical trials in OBC in women, with recommendations of mastectomy and axillary lymph node resection (8). The National Comprehensive Cancer Network guidelines recommend either mastectomy with axillary lymph node resection or axillary lymph node resection with whole breast irradiation for T0, N1, and M0 stage, and systemic chemotherapy, endocrine therapy, and combined surgery for stage II and III disease (9). However, there is no difference in survival rates between patients who undergo mastectomy and those who preserve their breasts, with adjuvant radiation being an effective treatment option (9). In addition, it is also deliberated that adjuvant chemotherapy and hormonal therapies are effective for OBC with node metastasis (10). Therefore, patients' preference and feasibility should be considered during clinical decision-making.

OBC with axillary metastasis is an extremely rare disease in men, with a high incidence of misdiagnosis. In this male patient, invasive ductal carcinoma from OBC was diagnosed in the axillary lymph node, even though the clinical and radiologic manifestations were similar to those of hematologic disorders, such as lymphoma or disseminated metastasis from unknown primary malignancies. In the future, multi-center studies are required to determine the appropriate treatment strategies for OBC in men and consequently, to improve clinical outcomes of this rare malignancy.

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상대 정맥 증후군으로 발현된 남성 잠재성 유방암의 증례 보고

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잠재성 유방암은 유방에 어떠한 증상없이 발생하는 유방암의 한 종류를 칭한다. 우리는 심한 호흡곤란과 상지부종을 보였던 72세 남성환자의 잠재성 유방암 증례를 보고한다. 전산화단층촬영에서는 양측 쇄골상 및 쇄골하 부위의 다수의 림프절 종대가 보였고 초음파 검사상 여러 개의 액와 림프절 종대가 발견되었다. 영상의학적 소견으로부터 임상적들은 림프종이나 불명의 원발성 암종의 파종성 전이를 의심했다. 액와 생검 표본으로부터 호르몬수용체 양성인 침윤성 유방암임이 밝혀졌다.

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