CASE REPORT

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Approaching Malignant Lymphedema: A Case Report

Min Gi Kim, MD, Jae Yong Jeon MD, PhD

Department of Rehabilitation Medicine, Asan Medical Center, University of Ulsan College of Medicine, Seoul, Republic of Korea

When managing cancer-related lymphedema, physicians should discriminate between benign and malignant types. Benign lymphedema could be solely treated by complex decongestive therapy. However, approach for malignant lymphedema should focus on the treatment of progressed cancer. Despite a lack of research on the assessment tools, identifying malignant lymphedema is important. We report a case of an intractable lymphedema patient whose lymphedema was previously assessed as benign. She was newly diagnosed with malignant lymphedema by computed tomography and showed significant improvement after chemoradiotherapy. This case report shows the clinical significance of CT venography for assessing malignant lymphedema in intractable lymphedema patients.

Key Words: Lymphedema · Malignant Lymphedema · CT Venography

INTRODUCTION

Lymphedema is the accumulation of protein-rich interstitial fluid developed by congenital or acquired lymphatic flow limitation due to surgery or radiotherapy-induced fibrosis[1]. Although the lymphedema may be benign, resulting from the impairment of lymphatic drainage system after radiotherapy or surgical cancer treatment, malignant lymphedema is also possible. In malignant lymphedema, malignant cells infiltrate the lymphatic system directly through recurrence or metastasis of the primary cancer[2]. Malignant lymphedema develops in 50% of patients with recurrent gynecologic cancer[3].

The treatment for malignant lymphedema differs from that of benign lymphedema. The standard treatment for lymphedema is complex decongestive therapy (CDT), which includes manual lymphatic drainage (MLD), bandaging, pump, exercise, and skincare[4]. However, MLD is limited in treating malignant lymphedema because it could cause aggravation by creating additional metastatic seeding[5,6]. In addition, differentiating between benign and malignant lymphedema is important to determine whether additional chemotherapy or radiotherapy is required[2].

To discriminate between the two types, there are various imaging tools for evaluation, such as ultrasound, computed tomography (CT), and positron emission tomography (PET). Fluorodeoxyglucose PET reportedly has good sensitivity for identifying malignant lymphedema;[7] however, research is lacking. Since the accessibility of PET is limited, additional studies on diagnostic methods are needed. Therefore, this case report aims to examine the progress of patients with malignant lymphedema confirmed by CT, which is relatively accessible.

CASE REPORT

A 54-year-old woman was diagnosed with cervical cancer by loop electrosurgical excision procedure con-

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Address for correspondence: **Jae Yong Jeon,** MD, PhD
Department of Rehabilitation Medicine, Asan Medical Center, University of Ulsan College of Medicine, Olympic-ro 43-gil 88, Songpa-gu, Seoul 05505, Republic of Korea
Tel: +82-10-9970-3791, Fax: +82-2-3010-6964
E-mail: jyjeon71@gmail.com

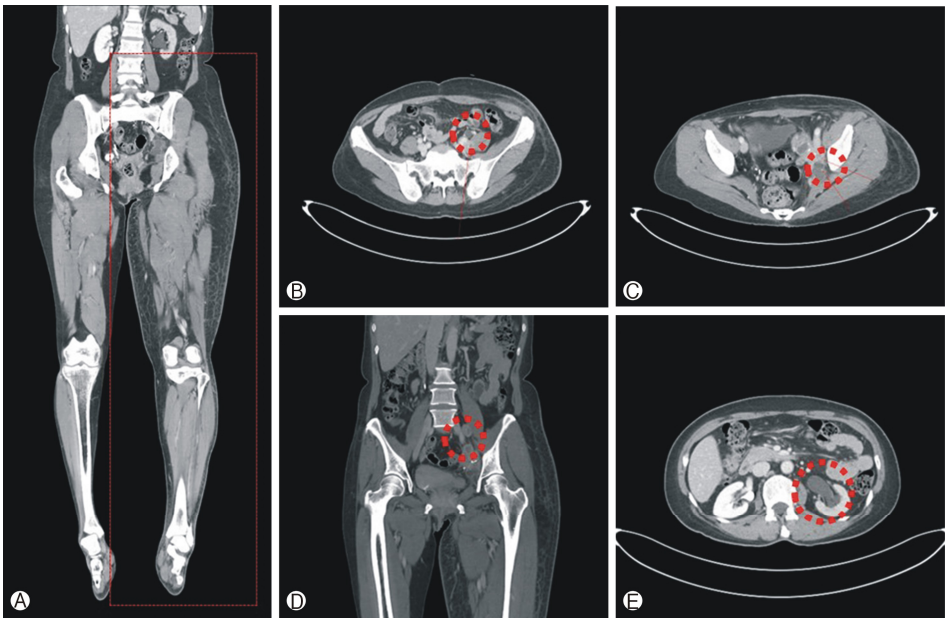


Fig. 1. Lower extremity CT venography of recurred cervical cancer patient with left persistent lymphedema.
This study was conducted for evaluating the persistent lymphedema. This images show disease progression; (**A**) diffuse left leg interstitial edema (**B**) solid and cystic mass along the left iliac vessels invading iliac lymph nodes (**C**) solid and cystic masses at left pelvic wall (**D**) Indistinct Lt mid-EIV area due to adjacent soft tissue masses (**E**) Lt hydronephrosis and hydroureter.

ization in 2002. She underwent a radical hysterectomy and bilateral pelvic lymph node dissection in the same year without adjunctive chemotherapy or radiotherapy. In May 2021, left lower limb edema developed and was confirmed as lymphedema by lymphoscintigraphy. Despite ongoing treatment for 5 months, including bandaging, there was no significant improvement. On October 22, 2021, she visited the Department of Rehabilitation Medicine of our hospital for further evaluation. Limb circumference was measured as 46.7/58.9 cm at 20 cm above the knee, 40.5/50.6 cm at 10 cm above the knee, and 30.5/36.7 cm at 10 cm below the knee. The patient's body weight was 58.7 kg at the time. The percentage excess circumference maximum (PEC max) was assessed as 26.1, and the ISL stage was 2.

On November 10, 2021, a recurrent mass was confirmed in the pelvic cavity, along with metastasis around the left iliac vessel and iliac lymph nodes. This confirmation was made through lower extremity CT venography, which was conducted to evaluate the presence of deep vein thrombosis and structural lesions (Fig. 1). On December 12, left percutaneous nephrostomy was performed due to left kidney complete obstruction on a renal scan. On December 15, torso PET scan revealed a hypermetabolic lesion in the pelvic cavity and the left common iliac area, including common iliac lymph nodes and lymphatic vessels. These findings were consistent with CT findings (Fig. 2).

On December 28, the PEC max was 30.4% at the outpatient clinic. She received thigh-high-compression stockings and educated bandaging of the lower limbs except for the inguinal area. A therapist could not routinely treat her due to the distance between the hospital and her home. She applied stockings during the daytime and bandaged during the night time. When bandaging was difficult, a garment was applied, and prophylactic antibiotics were provided in case of infection. Body weight was maintained around 57 kg with in-

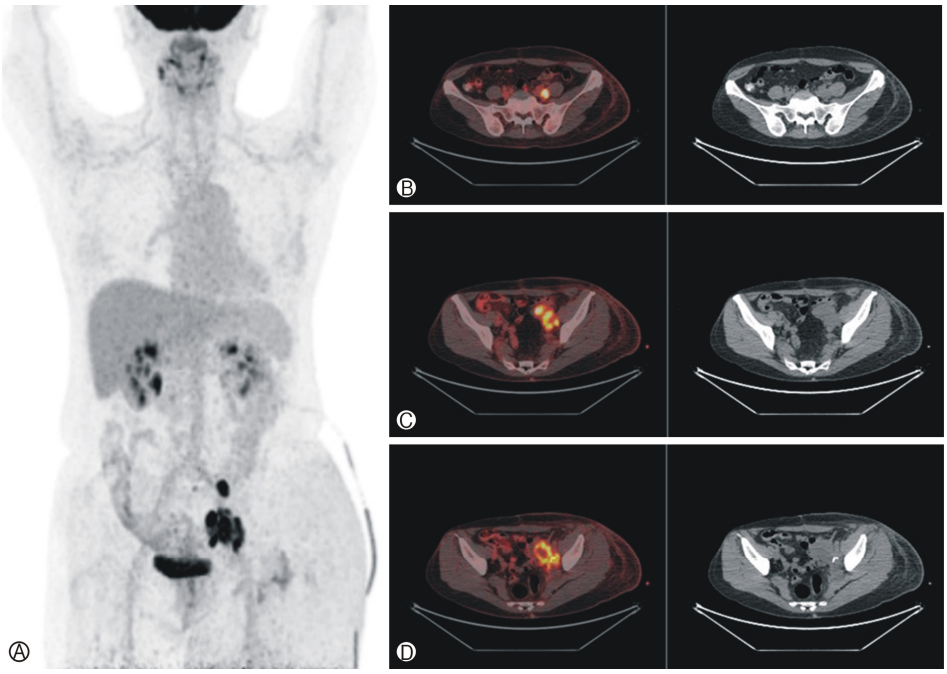


Fig. 2. Torso PET-CT of recurred cervical cancer patient with left persistent lymphedema. The patient underwent this study after CT scan. The images show (**A**) hypermetabolic mass and nodular lesions in left pelvic area (**B**) probable metastatic lymph node in left common iliac area (**C**, **D**) probable recurred mass around left pelvic wall.

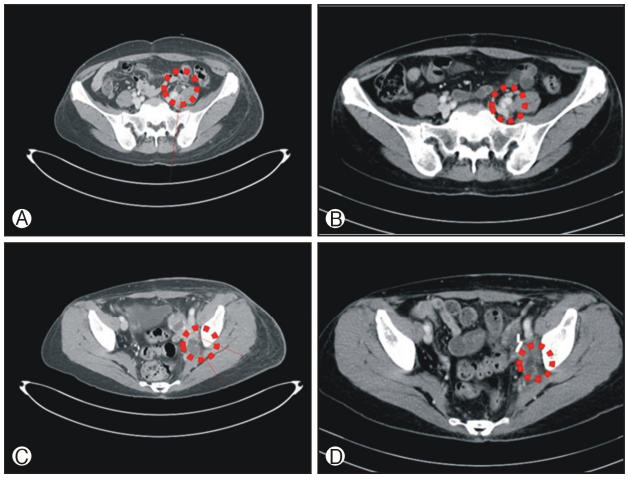


Fig. 3. Follow up CT scan of recurred cervical cancer patient with left persistent lymphedema.
CT scans (**A**, **C**) taken at the time of recurrence confirmation and CT scans (**B**, **D**) performed 4 months later for follow-up study were compared. During this period, chemotherapy and radiotherapy treatments were completed. These images show (**A**, **B**) improvement in the left iliac lymph nodes lesion and (**C**, **D**) a decrease in size was observed in the left pelvic wall lesion.

creased activity level and diet adjustment. Topical NSAIDs and oral medication were used for pain control.

From December 31, 2021 to February 18, 2022, six rounds of chemotherapy, including neoplatin, were conducted. From January 7 to February 25, extended field radiotherapy with a total of 33 fractions was irradiated with a total radiation dose of 66 Gy. The mass size has decreased on follow-up CT (Fig. 3). After the cancer-specific treatment, a dramatic improvement was observed, as PEC max 10.7%, when evaluated on July 8. However, due to the rapid aggravation on September 14, a severely compressed left mid-external iliac vein due to surrounding soft tissue (suggesting tumor recurrence) was identified on CT venography performed on September 29, and chemotherapy is being considered by the Gynecologist (Fig. 4).

DISCUSSION

Lymphedema can be categorized into primary and

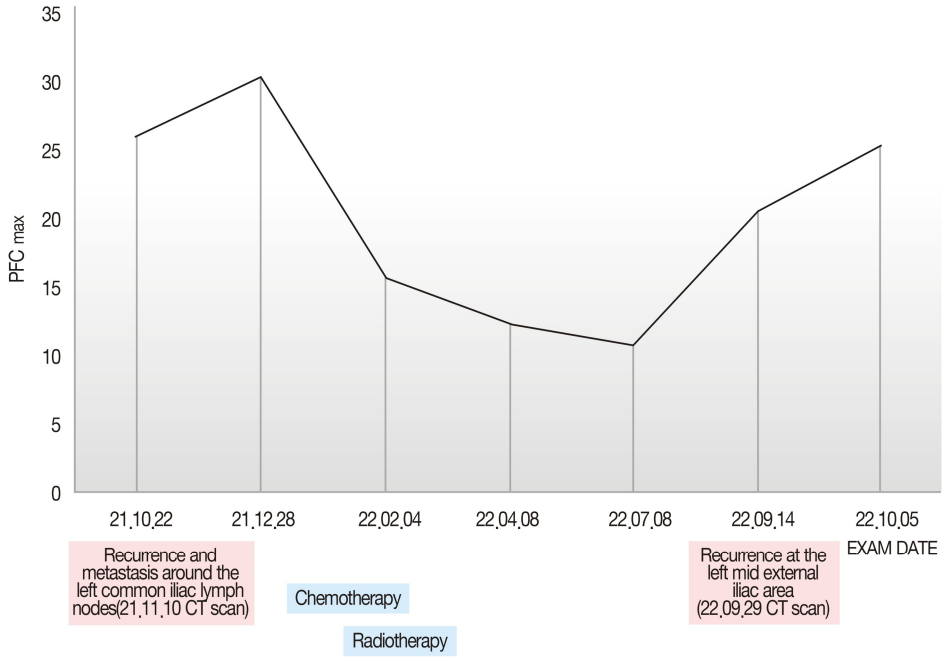


Fig. 4. Changes of left lower limb malignant lymphedema status.
The aggravation of lymphedema and tumor burden in late 2021. As chemotherapy and radiotherapy were initiated, PEC max was reduced. In September 2022, lymphedema aggravated along with the tumor burden.

Abbreviation: PEC max, percentage excess circumference maximum; CT, computed tomography.

secondary, which are relatively common in children and adults, respectively. The most common etiology worldwide is a parasitic infection, but lymphedema after treatment for malignancy is most common in developed countries[4]. Many clinicians might consider cancer-related lymphedema as a single disease entity. However, discrimination between benign and malignant types is essential owing to the different treatment trajectories. Furthermore, secondary malignant lymphedema, a variant of malignant lymphedema, can develop through indirect compression of the vessel by encircling a metastatic mass[8]. The patient in this case also had malignant lymphedema due to lymph node metastasis, but there was also a component of secondary malignant lymphedema because there was compression due to the recurred mass near the iliac vessel. However, distinction between these two subtypes does not alter the treatment.

In this case, malignant lymphedema due to tumor progression was confirmed on CT venography because there was no significant improvement despite the application of CDT for cancer-related lymphedema. Thereafter, lymphedema dramatically improved following a good response to chemotherapy and radiotherapy, however, worsened again despite continuing the lymphedema rehabilitation program. Another CT venography showed suspicious findings of disease progression, thus, additional chemotherapy is being considered. As such, ensuring an accurate diagnosis is essential because tumor-specific treatment is more important than lymphedema-specific treatment in malignant lymphedema.

Despite the limited studies on malignant lymphedema, there are several known characteristics; rapid and intractable progression, skin color change, accompanied by general weakness and unexplained weight loss[9,10]. Additionally, as observed in this case, lymphedema severity may reflect the tumor burden. Particularly in this population, the clinician should prescribe an imaging

modality to assess malignant lymphedema. Until now, PET is known to be the most sensitive tool, however, since sufficient anatomical information can be obtained with the more accessible CT, this tool should be considered first in clinical practice.

CONCLUSION

When approaching an intractable lymphedema patient, logical questionning of why CDT is not effective is important. Even in the typical clinical setting, clinicians should not hesitate to perform CT venography to evaluate the presence of malignant lymphedema and deep vein thrombosis. If malignant lymphedema is confirmed, fundamental cancer treatment should be performed first rather than the edema treatment.

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