Radiology Case Reports 18 (2023) 3041–3045

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journal homepage: www.elsevier.com/locate/radcr

Case Report

Lymphoscintigraphy with single-photon emission computerized tomography/computed tomography for evaluating lymphatic leakage following pelvic and para-aortic lymphadenectomy ☆,☆☆

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ARTICLE INFO

Article history:
Received 26 May 2023
Revised 4 June 2023
Accepted 8 June 2023

Keywords:
Interventional radiology
Lymphatic ascites
Lymphorrhea
Lymphoscintigraphy
SPECT/CT

ABSTRACT

Lymphatic ascites following pelvic and para-aortic lymphadenectomy is a well-known complication. Surgical treatment and interventional radiology are required in a few cases. To determine the appropriate treatment strategy, it is important to preoperatively detect the presence and location of lymphatic leakage. However, the methods have yet to be established.

We report a case in which lymphoscintigraphy with single-photon emission computerized tomography/computed tomography (SPECT/CT) was performed to evaluate pelvic lymphorrhea that occurred following total hysterectomy with pelvic and para-aortic lymphadenectomy for stage IIIA uterine sarcoma. Lymphoscintigraphy with SPECT/CT showed leakage of radioisotopes into the pelvic space, and intranodal lymphangiography was performed based on these findings. Following the procedure, the pelvic lymphorrhea improved, and no radioisotope leakage was confirmed by re-evaluation with lymphoscintigraphy with SPECT/CT.

Our case indicates that lymphoscintigraphy with SPECT/CT may be useful for detecting the precise site of lymphatic leakage before interventional radiology or surgery.

☆ Acknowledgments: I would like to thank Editage (www.editage.com) for English language editing.

☆☆ Competing Interests: The authors have declared that no competing interests exist.

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https://doi.org/10.1016/j.radcr.2023.06.023

Introduction

Lymphatic ascites following pelvic and para-aortic lymphadenectomy is a rare complication with a reported incidence of 1.0%-4.0% [1]. Most cases improve with conservative management, including drainage of the lymphatic effusion, total parenteral nutrition, and a low-fat diet containing medium-chain triglycerides [2]. However, a few cases are refractory to conservative therapy. In such cases, surgery or interventional radiology (IR) can be considered [3,4]. To determine the appropriate treatment strategy, it is important to preoperatively detect the presence and location of lymphatic leakage.

Several methods have been reported to determine the leakage point [5–7]. Lymphoscintigraphy is performed to evaluate the lymphatic system and is frequently used to diagnose patients with suspected cancer-related extremity lymphedema and to evaluate its severity. Some reports have indicated that lymphoscintigraphy may be helpful in diagnosing postsurgical lymphatic leakage in patients with lymphatic ascites. However, lymphoscintigraphy alone has low resolution, and it is difficult to identify the precise and detailed site of lymphatic leakage [8].

Single-photon emission computerized tomography (SPECT) is a nuclear medicine imaging technique that uses radioisotopes as well as lymphoscintigraphy, and is capable of providing 3-dimensional information. Moreover, the combination of SPECT and CT (SPECT/CT) allows for more precise detection of the anatomical location of radioisotopes than lymphoscintigraphy. However, the utility of SPECT/CT in detecting leakage sites in patients with lymphatic ascites has not been reported. We present a case in which lymphoscintigraphy with SPECT/CT was useful for the evaluation of pelvic lymphorrhea that occurred following pelvic and para-aortic lymphadenectomy with hysterectomy.

Case presentation

A 47-year-old woman underwent total hysterectomy with bilateral salpingo-oophorectomy, partial omentectomy, and pelvic and para-aortic lymphadenectomy for stage IIIA uterine sarcoma, according to the International Federation of Gynecology and Obstetrics classification. The surgery was uneventful, and the patient was discharged on postoperative day 6 and scheduled to receive adjuvant treatment with adriamycin. Two days after discharge, the patient visited our hospital because of abdominal fullness. Abdominal ultrasonography revealed abundant free abdominal fluid. The patient was consequently admitted to our hospital and underwent drainage with a percutaneous catheter with 8 liter volume drained on the first day. The fluid was colorless and transparent, and the analysis revealed a total protein concentration of 12 g/L, negative Rivalta test results, and no evidence of neoplasm. On the basis of these tests, the patient was diagnosed with postoperative lymphatic ascites. Lymphoscintigraphy was performed to diagnose lymphedema because the circumference of the patient's lower extremities had gradually increased. It was performed by subcutaneous injection of 40 MBq of 99mTc-labeled human serum albumin into the first and fourth web spaces of both feet. A planar image taken 90 minutes after injection showed a reduced number of inguinal lymph nodes accumulating isotopes bilaterally and abnormal extranodal isotope distribution in the pelvis (Fig. 1A). SPECT/CT demonstrated a hotspot in the pelvis on the right side, 2 cm above the superior margin of the femoral head (Fig. 1B–D).

The patient was managed conservatively with outpatient percutaneous and/or transvaginal catheter drainage once or twice weekly, and a low-fat diet with medium-chain triglycerides for 2 months. Despite conservative therapies, the volume of ascites remained at 4-6 liter per week.

The patient was scheduled for intranodal lymphangiography and percutaneous embolization of lymphatic leakage due to ascites refractory to conservative therapy. Ultrasound-guided access to the right inguinal lymph nodes was achieved using a 23G needle, and Lipiodol (Lipiodol Ultra-Fluid; Guerbet, Roissy, France) was administered. Fluoroscopic imaging demonstrated accumulation of Lipiodol in the right pelvis, which was also the hotspot site on lymphoscintigraphy with SPECT/CT (Fig. 2A).

Following the same procedure, intranodal lymphangiography using Lipiodol was performed at the left inguinal lymph nodes, which demonstrated normal lymphatic ducts from the inguinal to the lateral iliac region with no lymphatic leakage. Following the injection of 6 mL of Lipiodol, a CT scan was performed, which showed that Lipiodol did not spread throughout the abdominal cavity and remained localized only in the right pelvic cavity (Fig. 2B). Based on these findings, we determined that the lymphorrhea had spontaneously improved and that even if there was minor lymphorrhea, it could not be detected on lymphangiography as embolization could be expected due to the embolization ability of Lipiodol itself. Therefore, percutaneous embolization of the lymphatic tract was not performed. The patient was discharged from the hospital the day after intranodal lymphangiography without complications. The patient remained asymptomatic and had no recurrence of ascites on ultrasound for 6 months. Lymphoscintigraphy and SPECT/CT performed 6 months after intranodal lymphangiography showed no extranodal accumulation of radioisotopes in the pelvis (Fig. 3) and no deterioration of the lower extremity lymphedema.

Discussion

We report a case in which lymphoscintigraphy with SPECT/CT was performed to evaluate lymphatic ascites that occurred following total hysterectomy with pelvic and para-aortic lymphadenectomy for uterine sarcoma. Lymphoscintigraphy with SPECT/CT showed radioisotope leakage into the pelvic space, and intranodal lymphangiography was performed based on these findings. Following this procedure, the lymphatic ascites improved. Lymphatic ascites is a rare but severe complication that can cause abdominal distension, dehydration, electrolyte disturbances, and poor dietary intake secondary to nutrient loss. IR is a less invasive option than surgery for the treatment of lymphatic ascites. Several studies have shown

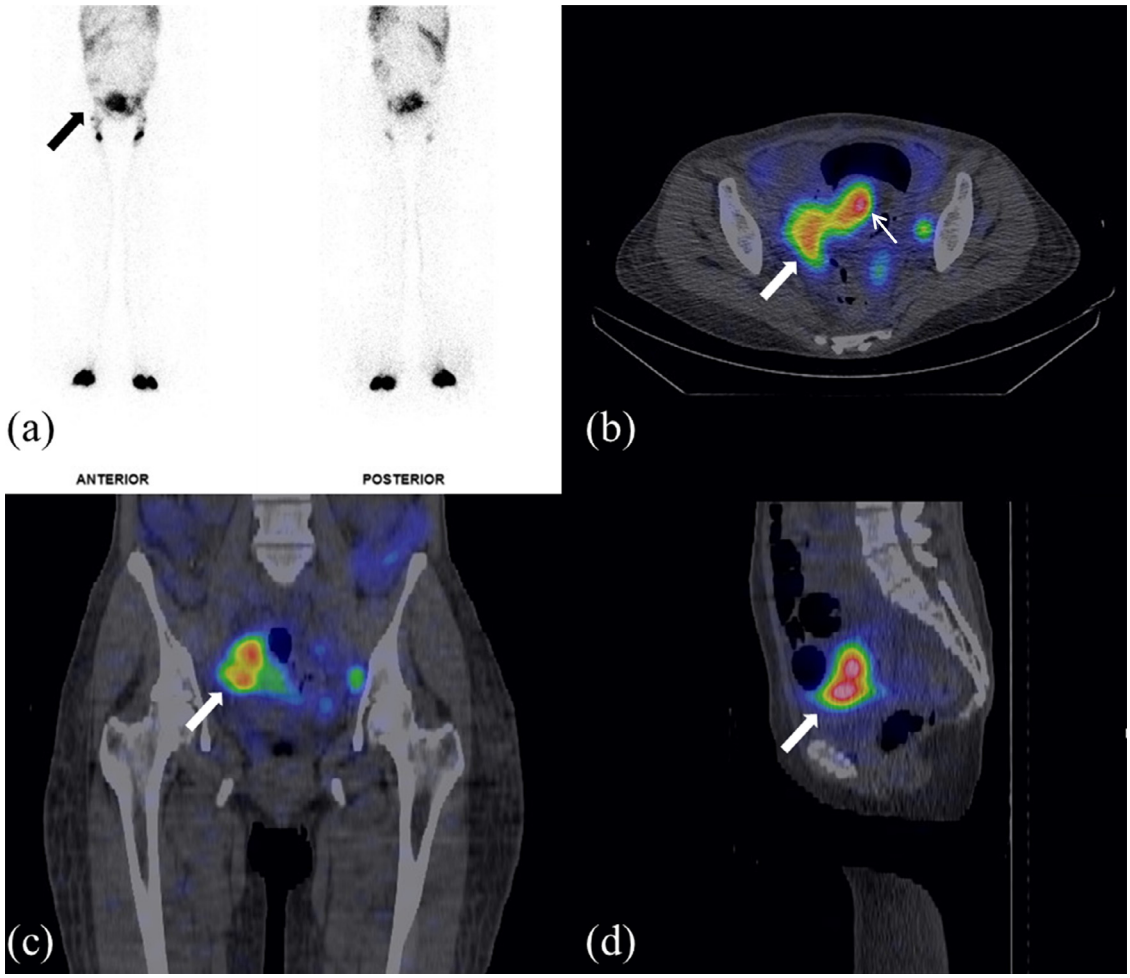

Fig. 1 – Planar image of lymphoscintigraphy amd multiplanar reconstruction of SPECT/CT. (A) Ninety minutes after the injection, showing the reduced number of inguinal lymph nodes accumulating the isotope bilaterally and abnormal extranodal isotope distribution in the pelvis (black arrow). Fusion axial (B), coronal (C), and sagittal (D) SPECT/CT images of the pelvis demonstrate the extranodal hotspot in the pelvic cavity on the right side, 2 cm above the superior margin of the femoral head (thick white arrow). On axial imaging, another hotspot is observed in the midline pelvic cavity (narrow white arrow). SPECT/CT: Single-photon emission computerized tomography/computed tomography.

that sclerotherapy and embolization are effective. Moreover, some reports indicate that lymphangiography alone using Lipiodol is effective for treating lymphatic ascites because Lipiodol causes inflammation and occlusion of lymph vessels, resulting in embolization of lymphatic leakage. The success rate of IR, however, depends on the amount of lymphatic leakage and varies in the literature, ranging from 35% to 83% [2,5,9,10].

Although surgical ligation is one of the most common physiologic treatments, it is sometimes difficult to identify the site of lymphatic leakage. Hence, a peritoneovenous shunt is performed instead of ligation when the site of leakage cannot be identified. However, this procedure is invasive, and can result in severe sepsis [3].

Several methods have been reported for detecting lymphatic leakage to achieve correct surgical ligation. As mentioned above, lymphangiography using Lipiodol is widely used for the preoperative evaluation of leakage sites [5,7]. However, it is invasive and requires skilled radiological techniques. Indocyanine green fluorescence imaging has also been reported as a useful intraoperative imaging technique for detecting leakage [6]. However, this method cannot provide preoperative information about leakage sites. Moreover, Lipiodol and indocyanine green are contraindicated in patients allergic to iodine.

Scintigraphy and SPECT/CT are used in functional imaging in various medical fields, including the evaluation of myocardial function, bone inflammation, cerebral blood flow, and lymphatic flow. SPECT/CT has a high resolution and can provide 3-dimensional information. Although some reports show that SPECT/CT demonstrates the site of minimal lymphatic leakage in patients with chylothorax and helps in occlusion by IR and surgery [11], the utility of SPECT/CT for detecting leakage sites in patients with lymphatic ascites has not been reported. In our case, we discovered the site of lymphatic leakage in a patient with lymphatic ascites after pelvic and paraaortic lymphadenectomy using SPECT/CT. The preoperative

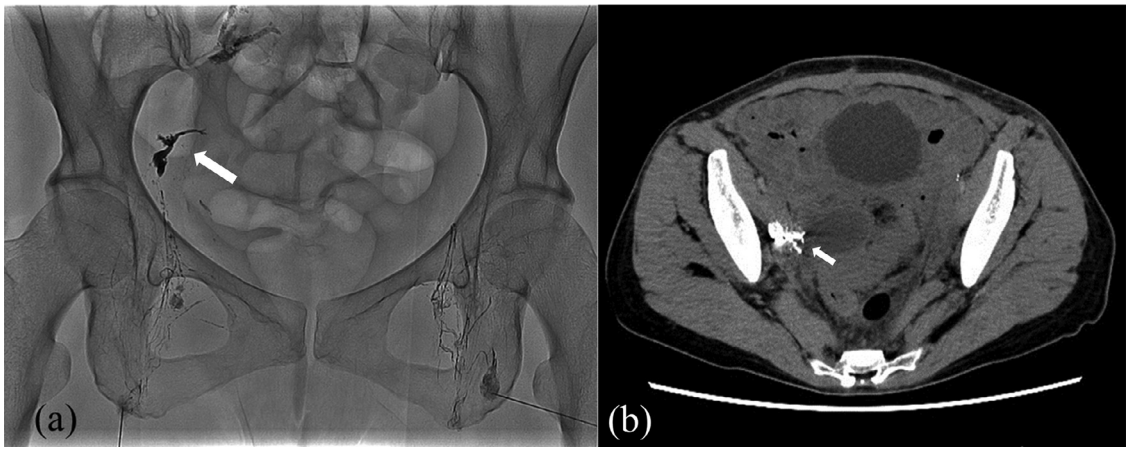

Fig. 2 – Fluoroscopic image and a computerized tomography scan during intranodal lymphangiography.
(A) Fluoroscopic image during intranodal lymphangiography. Lipiodol accumulated in the right pelvis (arrow), which showed a hotspot on single-photon emission computerized tomography/computed tomography. (B) A computerized tomography scan during intranodal lymphangiography. Extranodal accumulations of Lipiodol are observed in the right pelvic cavity but do not spread throughout the abdominal cavity (arrow).

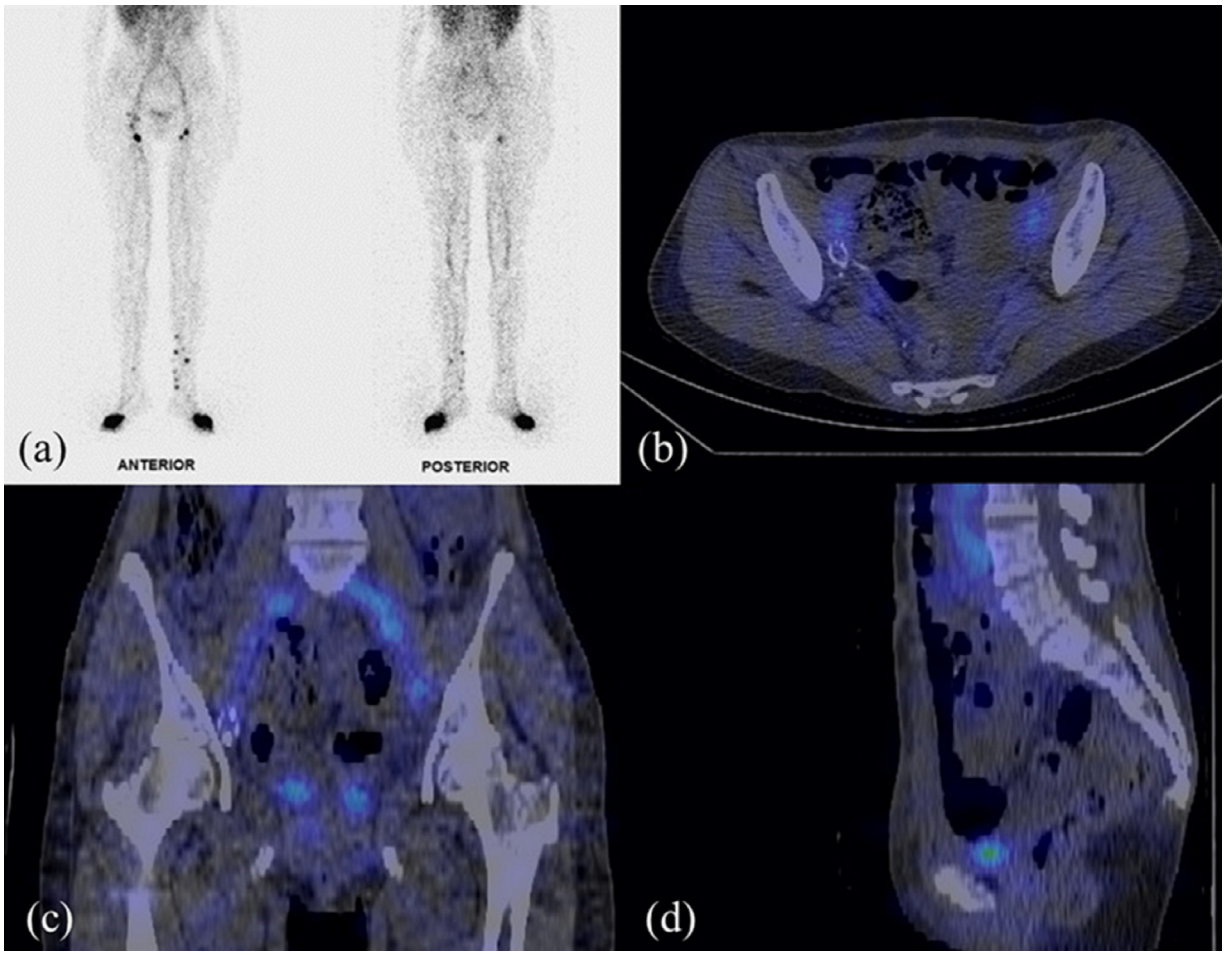

Fig. 3 – Planar image of lymphoscintigraphy and multiplanar reconstruction of SPECT/CT. (A) Six months after intranodal lymphangiography shows no abnormal extranodal isotope distribution in the pelvis. Fusion axial (B), coronal (C) and sagittal (D) SPECT/CT images of the pelvis also demonstrate no extranodal isotope distribution in the pelvic cavity. SPECT/CT: Single-photon emission computerized tomography/computed tomography.

information on the leakage site identified on SPECT/CT corresponded to the site of leakage found on lymphangiography using Lipiodol, which was successfully occluded by IR. This shows that SPECT/CT can facilitate safe correction of lymphatic leakage by surgery and IR.

Furthermore, SPECT/CT noninvasively showed reduced lymphatic leakage after IR in our case. Although improvement of lymphatic leakage is usually determined by CT and abdominal echograph [5,7], these examinations only evaluate the existence of ascites. SPECT/CT can be used to evaluate the existence of leakage both functionally and noninvasively.

Our study has some limitations. The sensitivity and specificity of SPECT/CT for detecting leakage sites are unknown because its use is still in the preclinical phase. In our case, SPECT/CT showed a hotspot where there was no accumulation of Lipiodol on lymphangiography (narrow arrow in Fig. 1B). This was considered to be the collection of leaked lymphatic fluid and a lymphatic cyst. The changes in lymphatic fluid over time should be monitored with SPECT/CT to distinguish them from leakage sites. If a hotspot is identified early, it could be a leakage site. In contrast, if the hotspot is identified only in the late phase, it is unlikely to be a leakage site. It is difficult to perform SPECT/CT over time. Hence, a suitable radioisotope tracer and time after injection for determining the leakage site need to be established in larger studies before routine use in clinical practice is recommended.

Our case indicates that lymphoscintigraphy with SPECT/CT may be useful for detecting the precise site of lymphatic leakage before IR or surgery. This can make the correction of lymphatic leakage by surgery and IR easier and more secure. Furthermore, SPECT/CT could be a useful postoperative examination to ensure improvement of leakage following noninvasive treatment.

Data statement

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

Ethical approval

Institutional review board approval was not required for this study. This study was considered to fall outside the scope of the Japanese legislation regulating research on human subjects, so the need for local ethics committee approval was waived.

Patient consent

Written informed consent for the publication was obtained from the patient, and anonymity was guaranteed.

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