



# Management of chylothorax after retrocrural lymphadenectomy in a patient with ovarian cancer: a case report

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**Introduction and importance:** With the widespread use of positron emission tomography and computed tomography (PET/CT), a significantly greater proportion of patients with advanced ovarian cancer (OC) are now diagnosed with superior renal-vein lymph node metastases involving retrocrural and mediastinal nodes. To the authors' knowledge, retrocrural lymphadenectomy has not yet been reported in patients with OC. The authors performed retrocrural lymph node resection in a patient with ovarian cancer.

**Case presentation:** A 64-year-old woman with ovarian cancer who had not undergone surgery upon initial diagnosis was admitted to the authors' hospital because tumour markers increased during bevacizumab maintenance therapy. PET/CT imaging revealed adnexal masses and multiple metastases in pelvic, para-aortic, retrocrural, and mediastinal lymph nodes. Reduction surgery was performed, and retrocrural lymph nodes were excised. However, the patient's postoperative course was complicated by a chylothorax. Because of the failure of conservative treatment, interventional embolization was performed, but failed to obstruct lymphatic vessels. The patient underwent reoperation. A fistula was located where Hem-o-lock clips penetrated the pleura, clearly indicating the injury site, which was then sutured and embedded in the surrounding diaphragmatic tissue and filled with gel sponge. The patient recovered from chylous leakage postoperatively. She later underwent chemotherapy and targeted maintenance therapy.

**Clinical discussion:** The authors may have injured the communicating branch of the thoracic duct posterior to the diaphragm during the first operation and did not ligate it. The accumulated chylous fluid finally penetrated through the weak point on the pleura and led to chylothorax 3 days later. If conservative treatment or interventional embolization are unsuccessful, surgical treatment should be selected in time.

**Conclusion:** The location of the retrocrural lymph node at the anastomosis of the chylous cistern and the thoracic duct may pose a significant risk of chylous leakage as a complication of lymphadenectomy. Full exposure of the surgical field and thorough ligation of the lymphatic vessels may lead to successful superior renal-vein lymphadenectomy.

**Keywords:** case report, chylothorax, ovarian cancer, retrocrural lymph node, ultraradical surgery

## Introduction

Ovarian cancer (OC) often metastasizes to multiple organs in the abdominal cavity. R0 resection is particularly important and closely related to prognosis. Lymphadenectomy is an important component of R0 surgery. The 2020 National Comprehensive

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## HIGHLIGHTS

- We report the first retrocrural lymphadenectomy indicated for ovarian cancer.
- Our patient developed the rare postoperative complication of chylothorax.
- Chylothorax failed conservative treatment but responded to surgical intervention.

Cancer Network Clinical Practice Guidelines in Oncology (NCCN guidelines) have recently revised the scope of ovarian cancer lymphadenectomy according to the Lymphadenectomy in Ovarian Neoplasms (LION) Trial<sup>[1,2]</sup>, and stipulate that enlarged pelvic and para-aortic lymph nodes in patients with stage IIB-IV OC must be surgically removed. If an enlarged lymph node is detected at the time of initial diagnosis, excision is recommended, even in cases in which the lymph node is successfully reduced after neoadjuvant chemotherapy.

Combined positron emission tomography and computed tomography (PET/CT) has been used successfully for the diagnosis of small lesions and distant lymphatic metastases<sup>[3,4]</sup>. Retrocrural (subdiaphragmatic) lymphadenopathy is often detected by PET/CT in the posterior diaphragmatic space, a triangular space between the posterior diaphragm foot and the lumbar vertebrae. Retrocrural lymphadenopathy is commonly

caused by tumours of the genitourinary, gastrointestinal, and respiratory tracts; inflammation; and infectious diseases. Retrocrural lymphadenectomy is rarely reported<sup>[5,6]</sup> and, to our knowledge, has not been described in the management of OC. Previous studies demonstrated that the incidence of chylothorax increased significantly with the resection of high-position para-aortic lymph nodes superior to the renal vein<sup>[4,7]</sup>.

Herein, we present a case of a 64-year-old woman with OC who underwent retrocrural lymphadenectomy and subsequently developed a chylothorax, which is a rare postoperative complication of lymph node resection indicated for metastatic OC<sup>[8,9]</sup>. We discuss the role of superior renal-vein lymphadenectomy in R0 surgery and the prevention of complications.

Ethical approval was obtained for this study. The patient provided informed consent for the publication of her clinical data and images.

### Case report

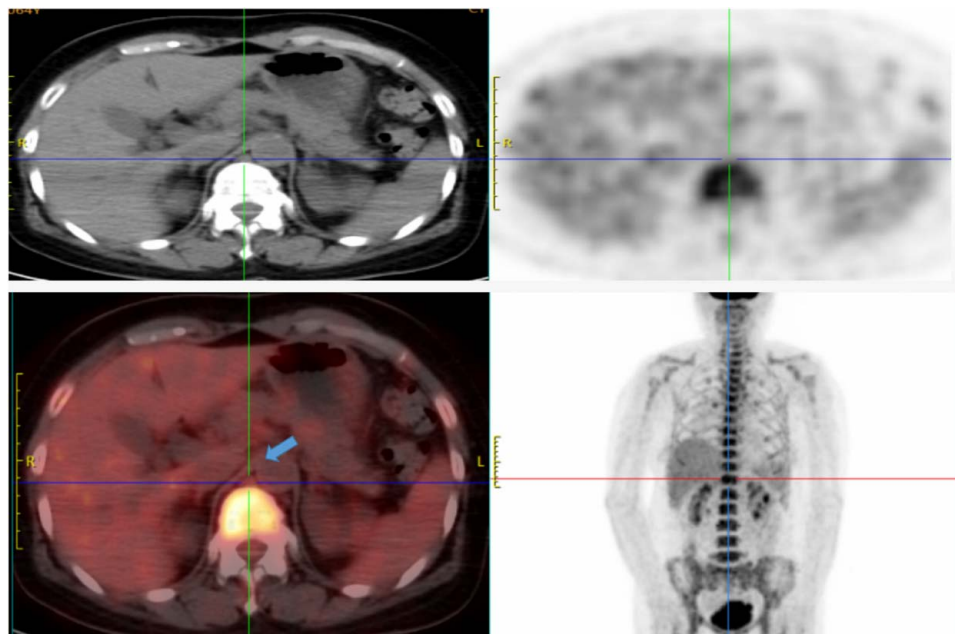
A 64-year-old woman was diagnosed with OC at another local hospital in July 2021. The patient had no family history of OC or breast cancer. PET/CT revealed a cystic and solid mass in the double appendages, and multiple pelvic, abdominal, and mediastinal lymph node metastases. The patient received five cycles of chemotherapy (paclitaxel liposomes combined with carboplatin) plus bevacizumab, but did not undergo surgery. Bevacizumab was administered as a maintenance treatment after chemotherapy.

In July 2022, the elevation of the tumour marker CA125 was noted and the patient was admitted to our hospital. The patient was in generally good condition. A pelvic mass was palpated during the gynaecological examination. PET/CT showed persistence of the ovarian tumour. Metastases to the right hilar, mediastinal, and right retrocrural lymph nodes were evident (Fig. 1).

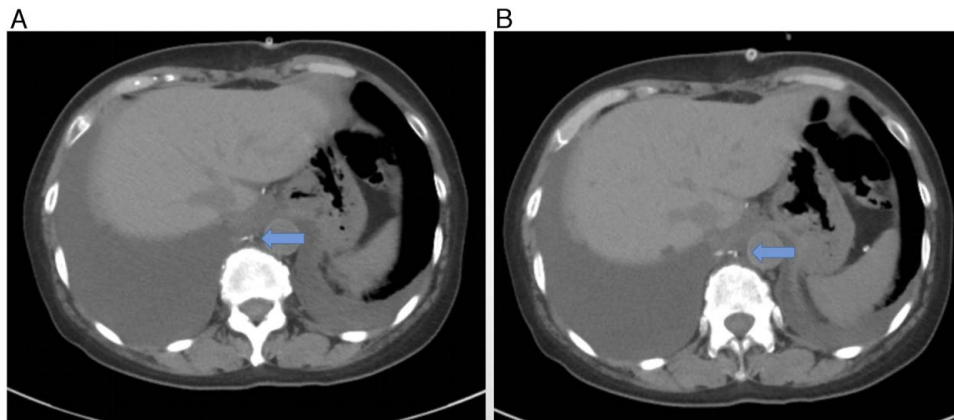
OC reduction surgery was performed by the attending doctor. The operation involved the uterus, double appendages, omentum, and appendix and included resections of pelvic and abdominal para-aortic lymph nodes and abdominal lesions. The right diaphragm was separated, and the retrocrural lymph nodes were resected. Revealed high-grade serous adenocarcinomas of both ovaries and multiple metastases to the pelvis and abdominal peritoneum. The regional lymph node metastasis status was as follows: left pelvic lymph node (0/1); right pelvic lymph nodes (3/4); subrenal vein lymph nodes (4/5); left gastric lymph nodes (1/1); and retrocrural lymph node (1/1). Partial intestinal obstruction and chylothorax developed postoperatively. Although the chylothorax was drained at a rate of 2000 ml/day, conservative treatments such as dietary restriction and octreotide application did not yield any notable effect. CT revealed that the Hem-o-lok clips were located under the diaphragm (Fig. 2A, B), indicating a potential site of chyloous leakage (Fig. 2A).

The patient received one postoperative cycle of paclitaxel combined with a carboplatin regimen. The chyloous leakage persisted, and the patient was referred to the interventional radiology department for embolization. A catheter was inserted into the thoracic duct through the right subclavian space, followed by retrograde placement into the cisterna chyli at the level of the first lumbar vertebra. Lymphangiography did not reveal an obvious fistula. The lower thoracic duct was occluded by using coils and adhesives (Fig. 3). After embolization, the drainage was slightly reduced, became turbid again, and finally increased to approximately 2000 ml/day. The positions of the spring coil and the adhesive are shown in Figure 3.

In October 2022, thoracotomy was performed after failed interventional therapy. During the surgery, we observed a fistula above the diaphragm with Hem-o-lok clips in its centre and surrounding exudates (Fig. 4A). Tissue of the caudal peripheral diaphragm was sutured using an absorbable thread and filled with gel sponge (Fig. 4B). The drainage fluid gradually decreased



**Figure 1.** Positron emission tomography and computed tomography images of the retrocrural lymph nodes. Increased metabolism is indicated by the arrow.



**Figure 2.** Computed tomography images showing the site of chylous leakage. (A, B) As indicated by the arrows, the Hem-o-lock clips were in the surgical site of retrocrural lymph nodes. (A) One Hem-o-lock clip was located just under the diaphragm, suggesting that this might be the site of chylous leakage.

and the thoracic drainage tube was removed. She returned to the hospital for chemotherapy and received maintenance treatment with bevacizumab. At present, there are no signs of recurrence, and she is tolerating treatment without complaints.

## Discussion

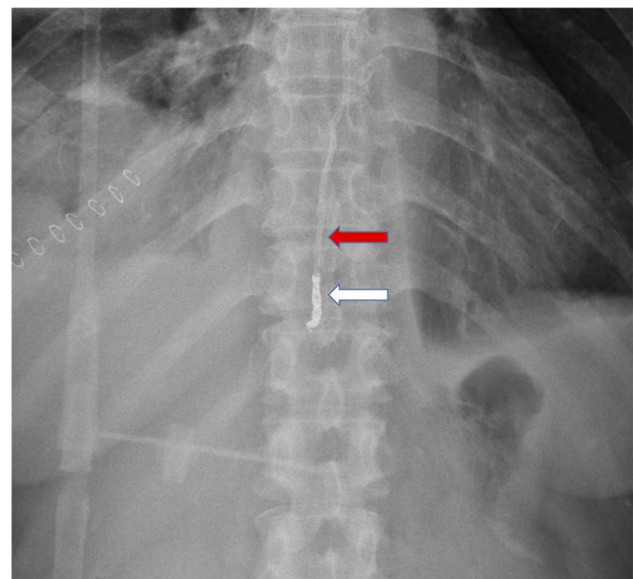
The introduction of PET/CT hybrid imaging in clinical settings has facilitated the identification of a higher incidence of superior renal-vein lymphatic metastasis. The increasing use and prognostic significance of R0 surgery have led experts to recommend R0 resection of all lymph node metastases. To our knowledge, retrocrural lymphadenectomy has not been reported previously in the management of OC patients. Herein, we present a case in which ultraradical surgery, including retrocrural lymphadenectomy, was performed successfully.

Retrocrural lymph nodes are located beneath the diaphragm. Several intestinal lymphatic vessels converge at the cisterna chyli above the renal vein. A retrocrural lymph node is located at the anastomosis of the chylous cistern and the thoracic duct. Therefore, in contrast to cardiophrenic lymph node resection<sup>[8–11]</sup>, retrocrural lymphadenectomy may easily traumatize the branches of the cisterna chyli and the thoracic duct, thereby predisposing to iatrogenic chylous leakage<sup>[7]</sup>. Although multiple Hem-o-lock clips were used in this case to clamp the lymphatic capillaries during resection, chylous leakage still occurred. Most chylous injuries cause leakage into the abdominal cavity. However, chylothorax can present 2–3 days after mediastinal injury<sup>[12]</sup>. In this case, the timing of the clinical presentation of chylothorax was consistent with that of other reported cases that followed predisposing injuries. The patient developed respiratory distress on the third postoperative day. CT revealed pleural effusion and finally led to the diagnosis of chylothorax. In retrospect, we may have injured the communicating branch of the thoracic duct posterior to the diaphragm and did not ligate it. The accumulated chylous fluid finally penetrated through the weak point of the pleura and led to chylothorax 3 days later.

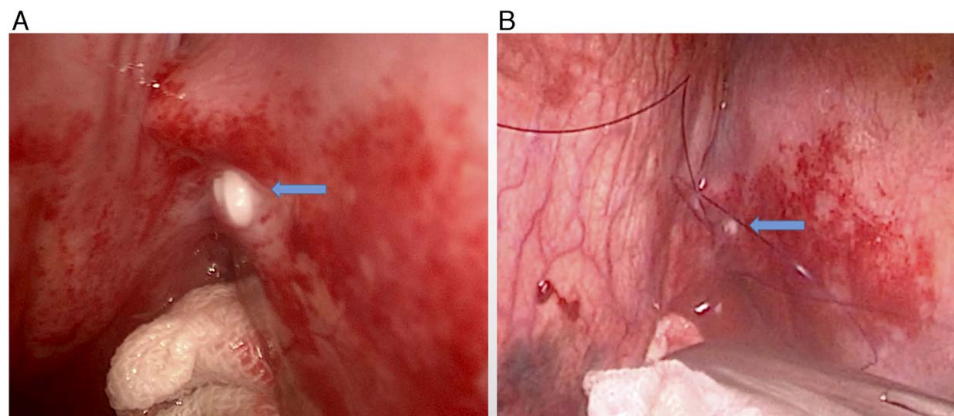
Postoperative chylothorax is more commonly observed in patients with oesophageal and lung cancer, and its management can be challenging<sup>[12,13]</sup>. Two OC patients who underwent

cardiophrenic angle lymphadenectomy presented with chylothorax after R0 surgery<sup>[8,9]</sup>. Conservative treatment, dietary restrictions, and octreotide administration may also be effective. Lymphography or surgery are recommended if conservative treatment is ineffective. In the present case, we performed angiographic surgery. The significant number of branches between the cisterna chyli and the thoracic duct impeded interventional embolization, and obstructing the trunk had no significant effect.

If interventional embolization fails to block lymphatic vessels, surgery is the only viable solution. The key to a successful operation is to locate the precise point of injury that may be identified during preoperative lymphangiography. A preoperative intestinal high-fat diet (cream or olive oil) may also facilitate the intraoperative identification of the exudation point. Surgical procedures should target dissociation and ligation of the upper thoracic duct at the injury site<sup>[7]</sup>. If the point of injury is



**Figure 3.** Chest X-ray after thoracic duct embolization. The coil and adhesive are indicated by the white and red arrows, respectively.



**Figure 4.** Suturing the chylous leakage site. (A, B) The fistula above the diaphragm, the Hem-o-lock clip indicated by the arrows, and exudate around the chylothorax are shown. (B) Tissue of the caudal peripheral diaphragmatic surface was sutured with absorbable thread.

unclear, clinicians should suture or clamp the tissue between the thoracic aorta and azygos vein or suture the upper area of the diaphragm. According to reports, identification and ligation of the perforated end of the thoracic duct may be safer but may be precluded by the presence of adhesions and inflammation.

In the present case study, Hem-o-lock clips placed during the first operation played an important role in identifying the injury site at the cisterna chyli on the CT scan. During the second operation, the fistula was identified where the Hem-o-lock clips penetrated the pleura, clearly indicating the injury point, which was then sutured and embedded in the surrounding diaphragmatic tissue and filled with a gel sponge.

Ultraradical surgery for OC with superior renal-vein lymph node metastasis requires further investigation. Therefore, the identification of an effective approach to systemic R0 resection with fewer complications is necessary. To prevent chylous leakage, full exposure of the surgical field and complete ligation of lymphatic vessels during retrocaval lymphadenectomy is necessary. Surgery should be performed when conservative treatment of chylothorax fails.

### Methods statement

This case has been reported in accordance with the SCARE criteria<sup>[14]</sup>.

### Ethical approval

It was approved by the Ethics Committee of the Affiliated Cancer Hospital of Shandong First Medical University. Number: SDTHEC2023006002.

### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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### Author contribution

L.C. and Q.W. designed the study. Y.H., F.G. and J.W. recruited the patients and collected the data. Q.W. and F.Z. Analyzed the data and prepared draft figures and tables. All authors were involved in writing the paper and provided final approval of the submitted and published versions.

### Conflicts of interest disclosure

The authors declare that there is no conflict of interest.

### Research registration unique identifying number (UIN)

Not applicable.

### Guarantor

Liang Chen.

### Data availability statement

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

**Provenance and peer review**

Not commissioned; externally peer-reviewed.

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