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Case Report

Overcoming therapeutic challenges in a case of refractory chylothorax following blunt abdominal trauma through the utility of cone-beam computed tomography lymphangiography

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Abstract: Chylothorax development following blunt abdominal trauma is a rare event. While low volume chylothoraxes usually improve with conservative treatment, invasive interventions are required in high-volume chylothoraxes with daily drainage exceeding 1000 ml. We report a case of a 14-year-old boy who developed high-volume (>3 L/d) chylothorax on the left side following blunt abdominal trauma. The patient did not respond to conservative treatments. He underwent thoracic duct ligation three times, with percutaneous lymphatic interventions, and percutaneous Lipiodol-glue mixture injections under computed tomography (CT) guidance, but all treatments were unsuccessful. We describe the four-month multidisciplinary challenge in this case that was ultimately successfully treated by surgical ligation of aberrant and/or collateral lymphatics draining from the left abdomen to the left pleural cavity, with the help of the novel cone-beam computed tomography (CBCT) lymphangiography (LAG) technique.

Keywords: Traumatic chylothorax, thoracic duct embolization, transvenous thoracic lymphangiography, cone-beam computed tomographic lymphangiography

Introduction

Chylothorax refers to the accumulation of chylous fluid in the pleural cavity due to thoracic duct injury [1-3]. Development of chylothorax following blunt abdominal trauma is an extremely rare and life-threatening condition [3-5]. Conservative treatment is generally unsuccessful in high-output chylothoraxe with daily drainage exceeding 1000 ml, and mortality rates greater than 50% are reported due to progressive malnutrition, immune suppression, and sepsis [3-6]. Thoracic duct mass ligation or thoracic duct and/or leak site embolization are therefore recommended in cases of high-volume leaks [1, 2, 7]. Thoracic duct mass ligation is also ineffective in the presence of collateral circulations or in cases of chylothorax with unusual presentations, as in our case [1, 8]. This report describes a patient who developed a left-sided high-volume chylothorax following blunt abdominal trauma and who was refractory to multiple thoracic duct surgeries and percutaneous lymphatic interventions over a four-month period. Successful treatment was ultimately achieved with cone-beam computed tomography (CBCT) lymphangiography (LAG). We encountered no previous case in which computed tomography (CT) and CBCT LAG were performed in addition to antegrade and retrograde lymphatic imaging and interventions in the pediatric age group.

Case report

A 14-year-old boy was admitted to the hospital with left flank pain that developed four days after a hard blow to the abdomen during a soccer match. A chest tube was inserted on the left side due to extensive pleural effusion. The pleural fluid had a milky appearance consistent with chylothorax, and his triglyceride level was 632

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Figure 1. Fluoroscopic image taken before lymphangiography shows a chest tube (arrow) in the left pleural space and multiple surgical clips (arrowheads) previously placed for thoracic duct ligation.

mg/dl. Despite conservative treatment, daily drainage ranged between 3200 and 4000 ml.

Thoracic duct ligation was performed 22 days post-trauma using video-assisted thoracoscopic surgery. A chest tube was inserted on the right side due to the effusion observed on that side (the first operation).

Thirty-one days after the trauma, the pleural drainage persisted, and thoracic duct ligation was performed over the diaphragm (the second operation) to treat lymphatic leakage possibly originating from the proximal thoracic duct or lymphatic collaterals related to the leakage. All possible sites around the aorta and esophagus over the diaphragmatic hiatus were ligated and clipped.

Forty-six days post-trauma, due to the continuing high-volume drainage in the left chest tube, intranodal LAG was attempted in order to identify the leak point, but was unsuccessful.

Sixty-four days after the trauma, the patient was referred to another center, where a laparotomy was performed under the diaphragm, together with ligation of the proximal thoracic duct up to the cisterna chyli. This third operation was also unsuccessful. The patient was then referred to our center, which has considerable experience in intranodal LAG and LAG-guided percutaneous interventions.

One hundred three days post-trauma, the patient underwent bilateral intranodal LAG under ultrasonography guidance in our center. Fluoroscopic images taken before LAG showed a chest tube (arrows) in the left pleural space and multiple surgical clips previously inserted for thoracic duct surgeries (**Figure 1**). The bilateral intranodal LAG procedure was carried out in line with the previously described standard technique [9]. Since the inguinal lymph nodes were used in the previous LAG, we were able to inject 5 ml of Lipiodol. The pelvic and intestinal lymphatics and cisterna chyli were poorly opacified. Sufficient information could not therefore be obtained regarding the location of the leak and abnormal lymphatics.

One hundred six days post-trauma, it was decided to image the thoracic duct by catheterizing it retrogradely at the left jugular-subclavian angle. A 5F introducer sheet was first installed in the left antecubital vein. The opening point of the thoracic duct into the vein was investigated at the jugular-subclavian angle using a 4F diagnostic catheter (**Figure 2A-E**). At the jugular-subclavian angle, the opening of the thoracic duct into the vein was empirically catheterized and visualized with water-soluble iodinated contrast agent. Under the road map, a 4F Uni-Select (USL) 2 diagnostic catheter was positioned at the venous opening of the thoracic duct, and a 2.7-Fr Progreat microcatheter was advanced through this under microwire guidance (**Figure 2B**). Due to the surgical clips placed on the thoracic duct in previous operations, we were unable to pass from the thoracic duct to the cisterna chili (**Figure 2C**). Only the distal part of the thoracic duct was therefore visualized by means of contrast agent (**Figure 2D**). Due to suspicion of extravasation, the thoracic duct was embolized with coils (**Figure 2E**).

Since no significant change was observed in the amount of drainage from the left chest tube in the following days, intranodal LAG was planned for a third time (**Figure 3A-D**). One hundred nine days post-trauma, previously unused lymph nodes were detected, and 8 ml of lipiodol was injected with a 25-gauge spinal nee-

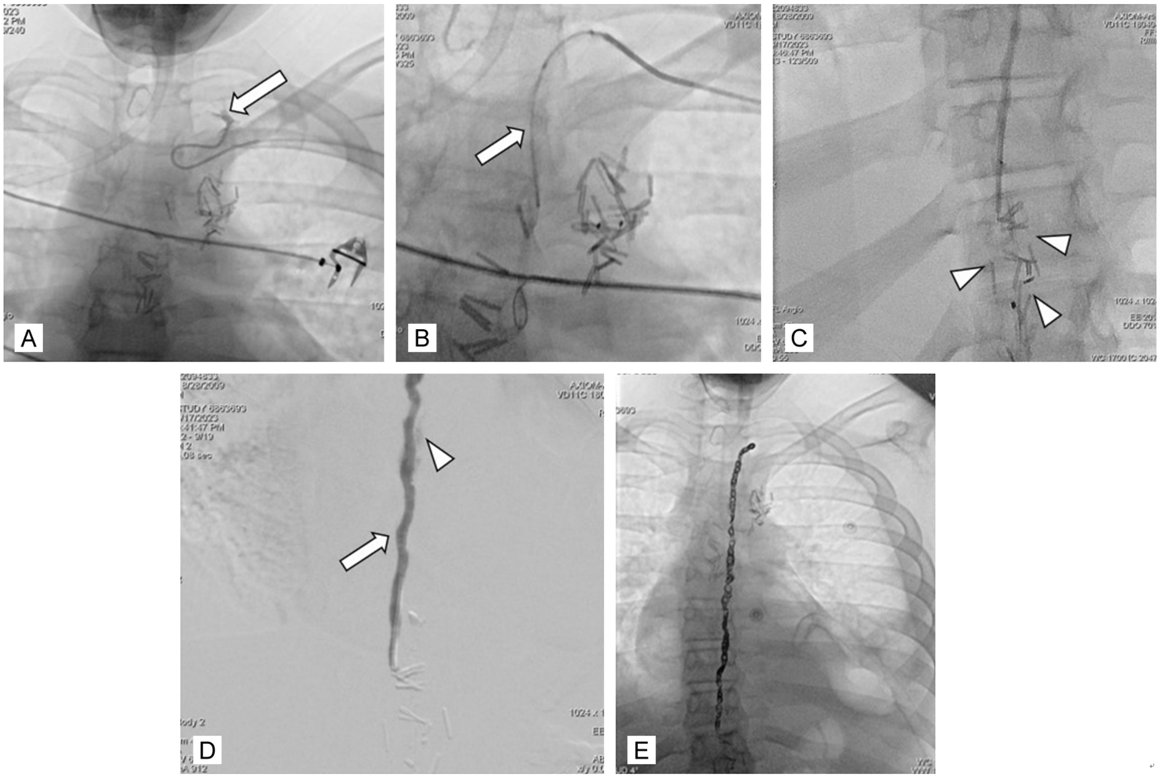

Figure 2. Terminal thoracic duct catheterization, imaging and coil embolization without thoracic duct opacification. A. Opacification of terminal thoracic duct (arrow) with a 4F Uni-Select (USL) 2 diagnostic catheter after catheterization of thoracic duct at the jugulosubclavian angle. B. After the thoracic duct was catheterized retrogradely, a 2.7-Fr Progreat microcatheter (arrow) was advanced through the diagnostic catheter. C. Due to the previously placed surgical clips (arrowheads), it was impossible to proceed to the proximal part of the thoracic duct and the cisterna chyli. D. Therefore, only the distal part of the thoracic duct was visualized (arrow) by administering a contrast agent. In addition, there is suspicious contrast extravasation (arrowhead) on the left side of the thoracic duct, which may be compatible with lymphatic leakage. E. The opacified segment of the thoracic duct was coiled to the point of opening into the subclavian vein.

dle under ultrasonography guidance. Twenty minutes after Lipiodol injection, the pelvic and abdominal lymphatics, and cisterna chyli were significantly opacified (**Figure 3A**). At this time, CBCT images were prepared from source images acquired by an Artis Q with Pure® biplane system (Siemens Healthcare GmbH, Forchheim, Germany) at the angiography unit. The cross-sectional images from Dyna CT were produced from 391 images (65 frames per sec) obtained during 200-degree rotation of the C-arm after processing on a Leonardo workstation. The acquisition time for the Dyna CT images was 6 s. The volumetric data were then reconstructed on the workstation to as axial-sagittal-coronal maximum intensity projections (MIPs) thin and 3D volume rendering technique (VRT) to determine the leakage side and level. Approximately 4-5 cm inferior to the cisterna chyli, multiple aberrant and/or collateral lymphatic channels originating from the left lumbar lymphatics were seen to be draining into the left hemithorax from the left side of the L1 vertebral corpus (**Figure 3A-C**).

Since surgical clips obliterated the proximal thoracic duct, the cisterna chyli could not be catheterized, despite being punctured percutaneously. A mixture of N-butyl cyanoacrylate (NBCA) and Lipiodol (NBCA:Lipiodol = 1:3) at a concentration of 25% in a volume of approximately 2 ml was therefore injected through a 21-gauge Chiba needle inserted percutaneously into the cisterna chyli (**Figure 3D**).

Although drainage decreased for a few days post-procedurally, it persisted during follow-up. One hundred fourteen days post-trauma, an NBCA-Lipiodol mixture at a 25% concentration (NBCA:lipiodol = 1:3) was injected into the aber-

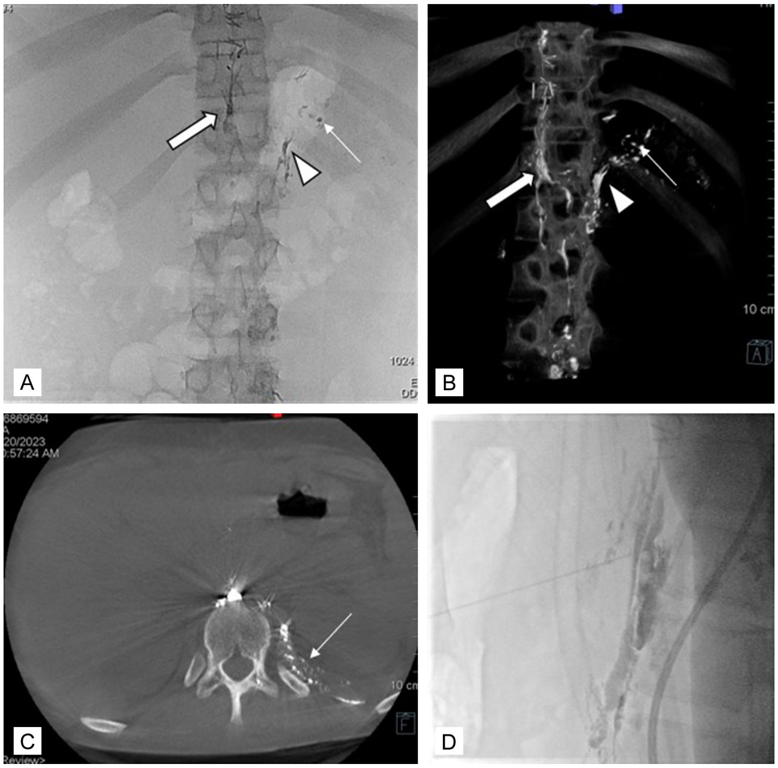

Figure 3. Antegrade bilateral intranodal lymphangiography from inguinal lymph nodes was tried again 3 days after thoracic duct retrograde embolization. A. Significant opacification of the cisterna chyli (arrow) at the T12 vertebrae level was seen 20 minutes after lipiodol injection from the lymph nodes in both groins. Aberrant collateral lymphatics (arrowhead) from the left lumbar lymphatics drain to the left hemithorax (thin arrow). B, C. Anatomic localization of aberrant collateral lymphatics (arrowhead), the cisterna chyli (arrow) and thoracic leakage (thin arrow) is better demonstrated in CBCT VRT and CBCT lymphangiographic source images. D. Lateral scopic image shows a mixture of lipiodol - N-butyl cyanoacrylate (NBCA) injection into the cisterna chyle with a 21-gauge Chiba needle.

rant lymphatics from two separate points under CT guidance (**Figure 4A**, **4B**). Post-LAG CT showed that the percutaneously injected lipiodol and NBCA particles had passed into the pleural space through aberrant collateral lymphatic channels. Although pleural drainage decreased to 900 ml per day in the 10-day period following the procedure, daily drainage persisted between 2000 and 2500 ml. The patient was operated for the fourth time in the light of detailed lymphatic anatomical information obtained from intranodal LAG, CBCT, and CT LAG at 121 days post-trauma.

All aberrant or collateral lymphatic ducts draining into the left pleural space on the left side of the aorta and L1-2 vertebrae were ligated through a left abdominal incision. Complete cessation of drainage was observed in the following 10 days, and normal oral feeding was initiated. The tube was withdrawn once the chest tube drainage decreased to 20 ml per day, and the patient was discharged with full recovery (**Figure 5A**). No recurrence of chylothorax was observed at 14-month follow-up (**Figure 5B**).

Discussion

This case illustrates the multidisciplinary challenges to diagnosis and treatment over a four-month period in a case of high-volume left chylothorax developing after blunting abdominal trauma. The first striking feature of this case is the unusual clinical presentation [1, 8]. Since LAG could not be performed initially, thoracic duct injury was theoretically considered as the cause of chylothorax, and all surgical operations were therefore performed empirically from different levels of the thoracic duct. Ligation of the thoracic duct distal to the leak site or collateral lymphatics seems to be the most important reason for the treatment failure and complicated the subsequent treatment.

The basic principle is that for effective lymphatic leak treatment, embolization or surgical ligation should be performed proximal to the leak site or abnormal lymphatics. Thoracic duct occlusions distal to the leak site raise intralymphatic pressure, which may lead to an increase in leak volume rather than leak closure, resulting in the opening of aberrant or new collateral lymphatic channels. When collateral lymphatics supply the leak site, as in our case, drainage continues even after ligation or embolization of the thoracic duct [1, 8-10].

There is currently no standard algorithm for the treatment of traumatic chylothorax. We created a diagnostic and therapeutic algorithm by sup-

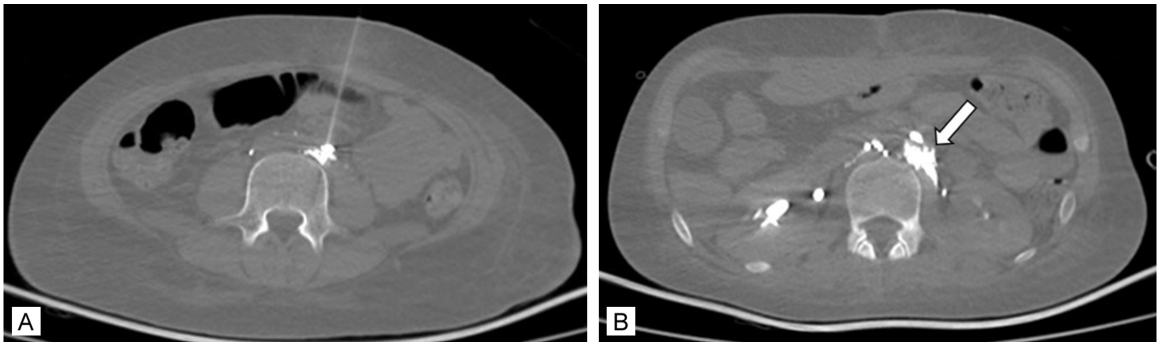

Figure 4. Under CT guidance, the left lumbar lymphatic duct inferior to the aberrant collateral lymphatics was accessed percutaneously with a 22-gauge Chiba needle. A. A mixture of NBCA) and lipiodol (NBCA:lipiodol = 1:3) was injected to into the left lumbar lymphatics below aberrant collateral lymphatics. B. Lipiodol-NBCA accumulation in and around collateral lymphatics after injection (arrow).

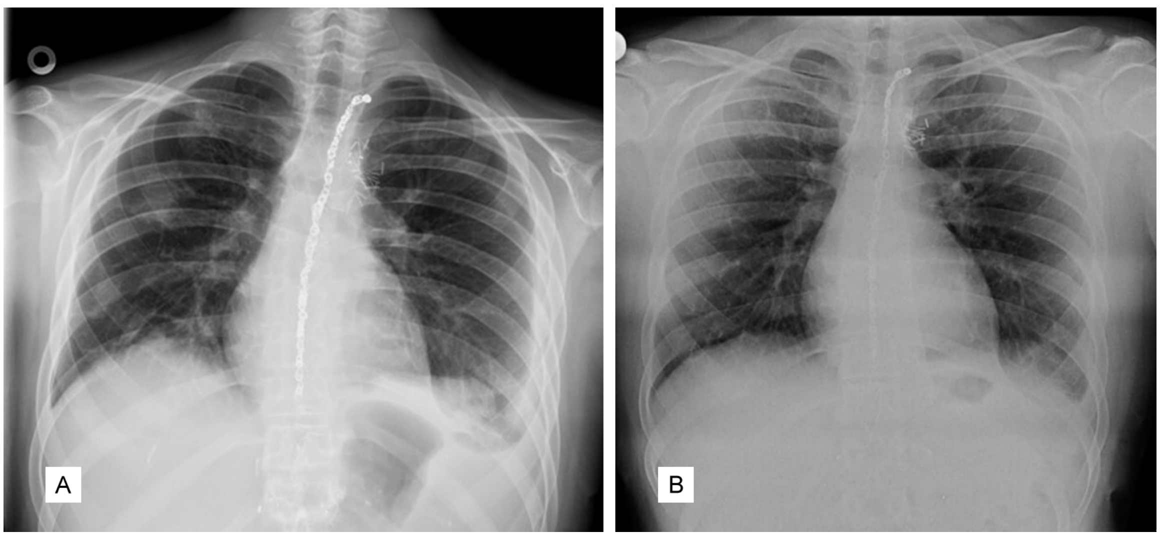

Figure 5. A PA chest radiograph on the day of discharge (A) shows metallic densities secondary to coils in the mediastinum and substantially decreased pleural effusion in the left hemithorax. The chest radiograph (B) taken 14 months later was normal except for metallic densities from the coils and surgical clips and blunting in the left costophrenic sinus which is a sequela of chylothorax.

porting our own experience with the findings of the previous literature (**Figure 6**) [6, 9-13]. The first step in the management of traumatic chylothorax is conservative treatment with diet modification and drainage of pleural effusion via chest tube or indwelling pleural catheter. The diet modification includes the use of medium-chain fatty acids that bypass the lymphatic system and are absorbed directly into the portal system, and some patients may require total parenteral nutrition. Treatment with Octreotide, a somatostatin analogue with an inhibitory effect on lymphatic fluid excretion, has also been shown to increase the effectiveness of conservative treatment. However, the efficacy of conservative treatment is lower than 50% in cases with chest tube drainage exceeding 500 ml per day [6, 11-14].

In cases unresponsive to conservative treatment, surgical thoracic duct ligation or interventional therapeutic options are applied. The first step in thoracic duct interventions is visualization of the thoracic duct and associated leakage with intranodal LAG. In low-volume (500 mL/d) lymphatic leaks, intranodal LAG alone can treat the majority (55%-65%) of leak sites. Numerous techniques can be used to treat the leak site with embolization. One of the most common of these is transabdominal per-

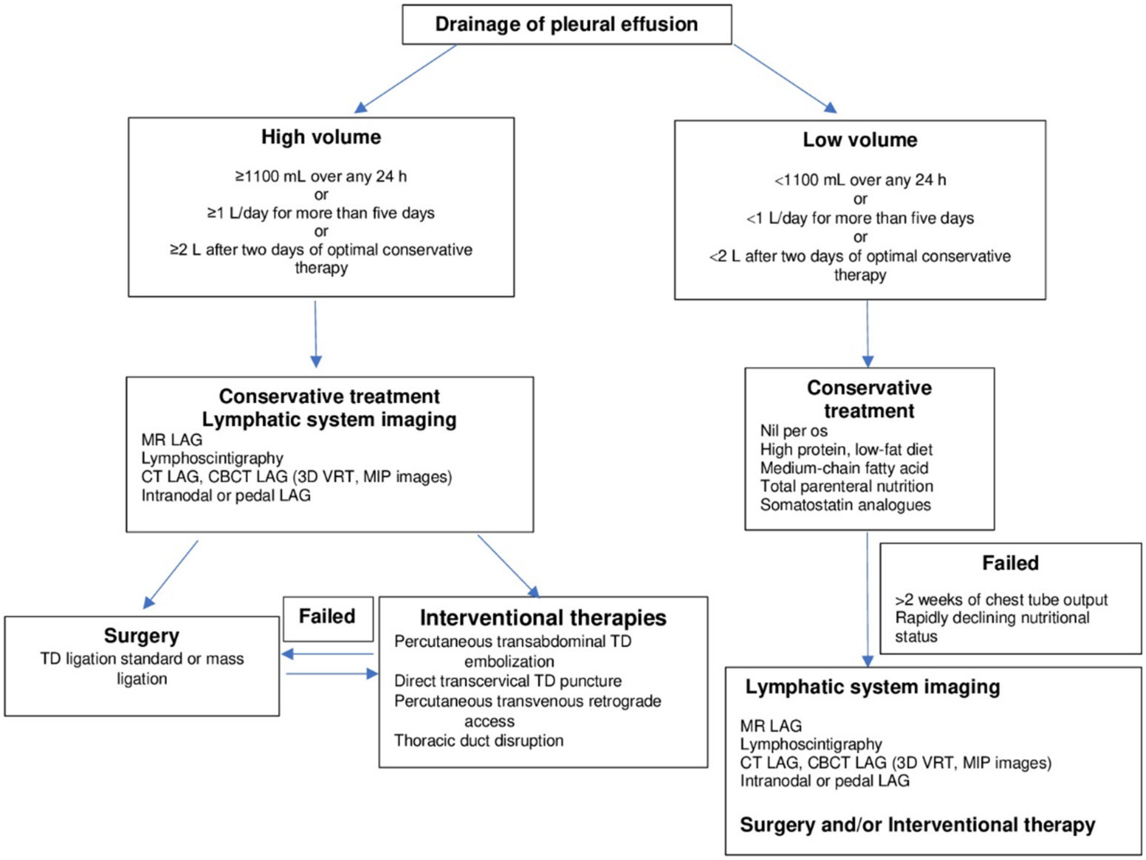

Figure 6. Proposed algorithm for management of traumatic chylothorax. MR LAG, magnetic resonance lymphangiography; CT LAG, computed tomography lymphangiography; CBCT LAG, cone-beam computed tomographic lymphangiography; 3D VRT, 3D volume rendering technique; MIP, maximum intensity projection; TD, thoracic duct.

cutaneous puncture of the cisterna chyli with a 21G needle, antegrade catheterization of the thoracic duct, and embolization of the leak site with a combination of coil and/or lipiodol-glue. The technical success rate for thoracic duct embolization varies between 57% and 100% [7, 9, 11-14].

Lymphatic leaks associated with collaterals after thoracic duct ligation may give rise to refractory chylothorax. Leaks associated with lymphatic collaterals are even more difficult to visualize and treat than direct thoracic duct-related leaks. We encountered a small number of case reports with similar characteristics to those of our patient in the literature [1, 8]. Kariya et al. [1] reported the difficulties encountered in the diagnosis and treatment of five cases of refractory chylothorax following esophageal cancer surgery. In three of these five patients, lymphangiography revealed extravasation of iodinated contrast medium in the collateral lymphatic duct, demonstrating a connection between the thoracic duct and the ruptured lymphatic duct via lymphatic collaterals. NBCA was used at very low concentrations (NBCA:Lipiodol = 1:5-1:20) for easier penetration of the embolizing agent because the lymphatic leaks associated with collaterals were far distant from the catheter tip. The NBCA-Lipiodol mixture penetrated to the point of lymphatic leakage in three patients. In the other two patients, thoracic ductography showed no direct communication between the thoracic duct and the ruptured lymphatic duct, and no contrast extravasation was detected. In these two patients, the NBCA embolized the cisterna chyle, lumbar trunks and lymphatic collaterals associated with the cisterna chyle. Embolization was successful in all cases. In our case, although NBCA was injected percutaneously into the cisterna chyle (**Figure 3D**) and under CT guidance (**Figure 4A**, **4B**) into the left lumbar lymphatics at the lower level of the collaterals, lymphatic leaks persisted. The most important reason for the failure of percutaneous interventions in our case was that the embolizing agent was unable to reach the collaterals and leakage area associated with the cisterna chyli and lumbar lymphatics due to the high NBCA concentration (NBCA:lipiodol = 1:3) used. Kariya et al. [1] therefore recommend the use of low concentrations of NBCA (NBCA:Lipoidol = 1/10-1/20) in cases of thoracic duct-related collateral leakage causing chylothorax.

Robinson at al. [8] reported a case of a post-operative high-volume, left-sided chylothorax refractory to all the usual recommended interventions. This was ultimately successfully treated by novel CT LAG - guided transabdominal surgical ligation of the aberrant left-sided lymphatics. Similarly in our case, precise anatomical location of the aberrant lymphatics draining into the left pleural cavity was performed with LAG and simultaneous CBCT LAG performed 103 days post-trauma, and definitive treatment was provided with abdominal surgery.

In addition to antegrade intranodal lymphangiography, the transvenous retrograde route can also be used as an alternative technique in imaging and interventional procedures in cases of chylothorax. The transvenous retrograde approach is generally recommended when the antegrade approach is not possible or when the cisterna chyle cannot be visualized. An important advantage of the transvenous retrograde approach is that potential complications of percutaneous cisterna chyle interventions are less common. The transvenous retrograde approach is usually performed in an antegrade manner after imaging the thoracic duct [9, 14, 15]. The original aspect of the transvenous retrograde imaging performed in our case was empirical catheterization of the thoracic duct in the left venous angle without antegrade thoracic duct visualization. In the present case, thoracic duct ligation performed distal to the leak site prevented us from reaching the leak site in a retrograde manner and treating it using the lymphatic pathway. If the thoracic duct had not been occluded with ligation, the leak site could have been easily accessed retrogradely, and the case could have been easily treated with a combination of coil and glue without the need for surgery.

The advantage of CBCT LAG is that it provides high resolution coronal, sagittal, axial, and oblique multiplanar images of the central lymphatic system without the patient needing to be removed from the table. The two-dimensional feature of conventional intranodal LAG may be insufficient to reveal the detailed anatomical structure of the lymphatic system in some cases due to superpositions. The anatomical details of lymphatic leaks detected at intranodal standard LAG can be better revealed by high-resolution three-dimensional images obtained from CBCT or computed tomography LG, which can be easily performed following intranodal LAG [16, 17]. In our case, detailed three-dimensional anatomical localization of the aberrant or collateral lymphatic channels associated with the left lumbar lymphatics by CBCT LAG ensured successful surgery. We encountered any other case in which CT and CBCT LAG were performed in addition to antegrade and retrograde lymphatic imaging and interventions in the pediatric age group.

In conclusion, this report describes a patient who developed high-volume chylothorax in the left hemithorax following blunt abdominal trauma and who underwent several surgical procedures, including intranodal LG, antegrade and retrograde lymphatic interventions, as well as CT LAG, CBCT LAG, and CT guided percutaneous interventions. It reveals the difficulties encountered and resolved through a multidisciplinary approach over a four-month period.

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Informed consent and agreement to publication both obtained from the family.

Disclosure of conflict of interest

None.

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