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ABOUT COVER

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CASE REPORT

Cholecystitis-an uncommon complication following thoracic duct embolization for chylothorax: A case report

Le Viet Dung, Ma Mai Hien, Thieu-Thi Tra My, Doan Tien Luu, Le Tuan Linh, Nguyen Minh Duc

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Abstract

BACKGROUND

Chylothorax is an uncommon condition in which chyle leaks into the pleural cavity, and biliary peritonitis is a rare complication of thoracic duct embolization in clinical practice.

CASE SUMMARY

We describe the case of a 50-year-old woman who presented with chylothorax and underwent thoracic duct embolization using a coil and a mixture of histoacryl glue and lipiodol. The patient developed upper abdominal pain and fever after the intervention. She was diagnosed with biliary peritonitis and treated with cholecystectomy at Hanoi Medical University Hospital.

CONCLUSION

Although thoracic duct embolization is considered a safe and minimally invasive procedure, it is not without risk. Following thoracic duct embolization, severe or persistent abdominal pain should be explored utilizing imaging data and laboratory results to determine problems as soon as possible.

Key Words: Lymphangiography; Thoracic duct embolization; Cholecystitis; Chyle leakage; Biliary peritonitis; Case report

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Core Tip: Chylothorax is a condition in which chyle leaks into the pleural cavity. Trauma, spontaneous (non-traumatic), and idiopathic etiologies are all potential causes of chylothorax. In clinical practice, biliary peritonitis is an uncommon consequence of thoracic duct embolization. Despite its reputation as a safe and minimally intrusive technique, thoracic duct embolization is not without hazard. Severe or chronic abdominal discomfort should be investigated as soon as possible after thoracic duct embolization, using imaging data and laboratory results to identify issues.

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INTRODUCTION

Chylothorax is a rare disease characterized by chyle leakage into the pleural cavity. The etiology of chylothorax can be traumatic, idiopathic, and non-traumatic including malignancies, primary lymphatic disorders or congenital malformations^[1]. Conservative management consisting of dietary control and drainage is typically the first-line treatment. Interventional radiology and surgery are considered if conservative management fails[1]. Thoracic duct embolization is a new, safe, and effective method for chylothorax treatment, it is associated with minimal complications [2,3], and these attributes have contributed to the recent increase in the use of thoracic duct embolization as a first-line treatment for chylothorax^[4]. In this case report, we describe the occurrence of biliary peritonitis following thoracic duct embolization for the treatment of chylothorax due to penetration of a percutaneous interventional needle into the gallbladder wall.

CASE PRESENTATION

Chief complaints

A 50-year-old woman presented at Hanoi Medical University Hospital. with dyspnea and chest pain.

History of present illness

The symptoms started 1 mo prior to presentation, with chest discomfort.

History of past illness

Medical records were normal.

Personal and family history

There was no personal and family history of other diseases.

Physical examination

Physical examination was not special. On auscultation, vesicular breath sound was diminished with decreased vocal resonance especially at the middle and lower lobes of the right lung.

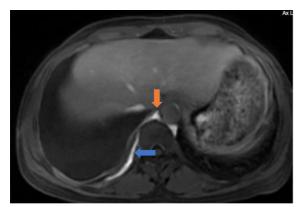
Laboratory examinations

Blood tests at presentation were in the normal range. Her pleural fluid analysis showed: Triglyceride 20.3 mmol/L and cholesterol 2.2 nmol/L.

Imaging examinations

Contrast-enhanced magnetic resonance (MR) lymphangiography was performed to obtain coronal and axial T1-weighted and T1-weighted fat-saturated post-gadolinium images, which showed a large right pleural effusion and contrast agent leakage from the right-sided branch of the lower-third of the thoracic duct into the right pleural space (Figure 1) and a double thoracic duct variant.

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Figure 1 Contrast-enhanced magnetic resonance (MR) lymphangiography before treatment. Axial T1-weighted fat-saturated post-gadolinium image revealed the leakage of contrast agent (blue arrow) from a right-sided branch of the thoracic duct (orange arrow) into the right pleural cavity.

INITIAL DIAGNOSIS

Further diagnostic work-up

A multidisciplinary consultation (involving respirologists, interventional radiologists, nutritionist) was performed. Three days after presentation, the patient underwent an intranodal lymphangiogram using digital subtraction angiography (DSA), which revealed dilated lymphatic vessels and lymphatic malformations in the lower-third of the thoracic duct, resulting in the leakage of contrast agent into the right pleural space (Figure 2). The patient also had a double lower thoracic duct variant. As the leakage was clear on MR lymphangiography and the patient had a large right pleural effusion (Figure 1), we decided to treatment her with lymphatic embolization, combined with lipid fasting. Under DSA guidance, the cisterna chyli was accessed percutaneously through the anterior abdominal wall with a 22-gauge Chiba needle. A right transabdominal approach was performed to avoid the aorta. The contrast was injected to confirm appropriate placement in the cisterna chyli. A wire was then advanced through the needle and placed within the thoracic duct. The thoracic duct below the leak was embolized under DSA, using a coil and a mixture of histoacryl glue and lipiodol.

Several hours after this intervention, the patient reported moderate-severe epigastric and right quadrant pain. Contrast-enhanced computed tomography (CT) was immediately performed, which showed wall thickening and subserosal edema in the gallbladder (Figure 3), with peripancreatic fluid extending into the pararenal spaces and bilateral paracolic gutters. The pancreas was homogeneously enhanced, with no signs of necrosis or pancreatic duct dilatation. A small, high-density nodule was observed in the gallbladder wall. The initial diagnosis was acute pancreatitis after lymphatic intervention; thus, she underwent conservative treatment instead of surgery. The respiratory symptoms were resolved by the intervention, but the patient experienced increasing abdominal pain and mild fever. Blood tests revealed an infectious condition: white blood cell count (WBC), 13.5 G/L; neutrophils, 88.7%; and C-reactive protein (CRP), 3.49 mg/dL. Pancreatic enzymes were within the normal range: amylase, 84 U/L; lipase, 20 U/L; and total/direct bilirubin, 9.6/5.7 µmol/L. A second contrastenhanced abdominal CT performed 3 days later revealed further gallbladder wall thickening (approximately 20 mm) and subserosal edema (Figure 4), with no signs of stones, pseudoaneurysm, or contrast agent leakage. The small, high-density nodule on the gallbladder wall and approximately 30 mm of ascites were observed.

FINAL DIAGNOSIS

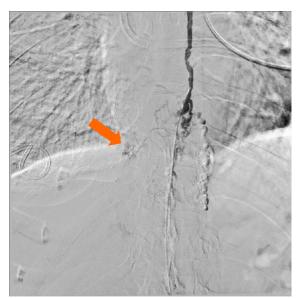
The final diagnosis was biliary peritonitis following thoracic duct embolization for chylothorax treatment. The critical finding was the high-density nodule observed on the gallbladder wall, which corresponded to lipiodol deposition following penetration of the percutaneous interventional needle into the gallbladder wall.

TREATMENT

The patient underwent emergency laparoscopic cholecystectomy. The surgeon described 300 mL of bile fluid in the abdominal cavity. The gallbladder was inflamed and covered with greater omentum. The surgeon diagnosed biliary peritonitis in this case. Two holes (approximately 1 mm) were identified in

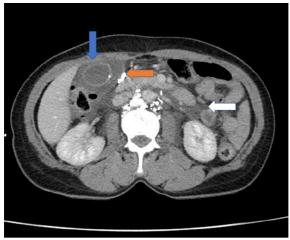


Dung LV et al. Iatrogenic cholecystitis following thoracic duct embolization



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Figure 2 Intranodal lymphangiogram using digital subtraction angiography prior to treatment. Frontal image of digital subtraction angiography revealed dilated lymphatic vessels and a lymphatic malformation in the lower thoracic duct (orange arrow), resulting in the leakage of contrast agent into the right pleural space.



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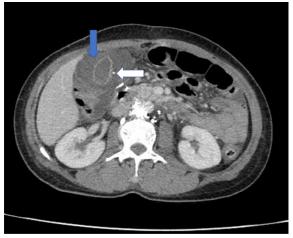
Figure 3 Contrast-enhanced computed tomography after intervention. Axial image showed gallbladder wall thickening of approximately 14 mm and subserosal edema (blue arrow). Peripancreatic fluid is observed extending into the pararenal spaces (white arrow) and bilateral paracolic gutters. A small, high-density nodule is observed in the gallbladder wall (orange arrow).

the base and funnel of the gallbladder (Figure 5), suggesting penetration of the gallbladder wall by the percutaneous interventional needle.

OUTCOME AND FOLLOW-UP

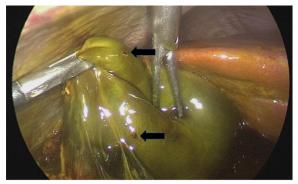
After surgery, the patient improved clinically, and the chylothorax output gradually resolved after several weeks. Inflammatory markers remained slightly elevated, and bilirubin returned to the normal range within 5 days: WBC, 9.3 G/L; neutrophils, 82%; CRP, 2.84 mg/dL; and total/direct bilirubin, $3.2/12.2 \mu$ mol/L. Abdominal ultrasound showed mild fat stranding around the gallbladder bed and 24 mm of right pleural effusion. The patient was discharged from the hospital 5 days after surgery.

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Figure 4 Contrast-enhanced abdominal computed tomography after 3 days. Axial image shows further increased gallbladder wall thickening (~20 mm) and subserosal edema (blue arrow), without evidence of stones, pseudoaneurysm, or contrast agent leakage. A small, high-density nodule can be observed in the gallbladder wall (white arrow).



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Figure 5 Laparoscopic cholecystectomy. Image revealed 2 holes 1 mm in size (black arrows) in the base and funnel of the gallbladder.

DISCUSSION

Lymphatic intervention was first described by Cope *et al*[5], and advances in interventional techniques continue to be developed. The efficacy of lymphatic intervention has been reported by many authors[2, 6,7]. Thoracic duct embolization is a new and minimally invasive approach with a high success rate and low morbidity and mortality rates^[2]. Cope and Kaiser reported a response rate of 73.8% after thoracic duct embolization with no morbidity or mortality[4]. The overall clinical success rate reported by Pamarthi *et al*^[3] was 72%, and Itkin *et al*^[7] reported a success rate of 90%.

According to the Society of Interventional Radiology Clinical Practice Guidelines, post-intervention complications can be categorized as major or minor complications[8]. Major complications of lymphatic interventions include pulmonary or portal vein embolism caused by glue or guidewire fragment retention. Minor complications often include anesthesia side effects, skin injuries, or perihepatic hematoma[3]. In addition, some delayed mild complications as observed by Laslett et al[9] are lowerextremity edema and chronic diarrhea.

Cope et al[4] reported no complications associated with thoracic duct embolization. Pamarthi et al[3] and Itkin et al[7] reported complication rates of 6.7% and 3%, respectively. Although limited data are available regarding complications, several articles have reported pulmonary embolisms associated with glue, chylous ascites, perihepatic hematoma, bile leakage, or bile peritonitis[3,10,11].

As chylothorax in this patient lasted for 1 mo, not an emergency condition, we decided to treat after a multidisciplinary consultation. Therefore, thoracic duct embolization was performed 3 days after hospitalization. Our patient experienced moderate-severe abdominal pain and fever following thoracic duct embolization, which increased over several days. A contrast-enhanced abdominal CT performed immediately after the intervention revealed that the gallbladder wall was thick and edematous, with peripancreatic fluid extending to the bilateral anterior pararenal space and paracolic gutters. The initial diagnosis was pancreatitis, which is also an extremely uncommon complication following thoracic duct



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embolization^[12]. Unfortunately, the clinical presentation, laboratory tests, and imaging with contrastenhanced abdominal CT 3 days later were not compatible with the initial diagnosis. We reviewed the initial CT images and realized that we missed a critical sign: a small, high-density nodule in the gallbladder wall, corresponding to lipiodol deposition due to penetration of the percutaneous interventional needle into the gallbladder wall. As the needle punctured through the right abdominal wall, it penetrated into the gallbladder. Although this is a rare complication infrequently reported in the literature, we suggest that the cisterna chyli should be accessed under DSA accompanied by ultrasound in order to avoid accidental puncture in the future. The patient had symptoms of peritonitis, and bile in the abdominal cavity was confirmed intraoperatively. Thus, the final diagnosis was biliary peritonitis. This case serves as a reminder for clinicians and radiologists of the importance of correct diagnosis following the initial CT scan to allow for timely cholecystectomy.

CONCLUSION

We report a rare case of cholecystitis following thoracic duct embolization to treat chylothorax due to penetration of the interventional needle into the gallbladder wall. Although thoracic duct embolization is considered to be a safe and minimally invasive method, several major complications may occur. Severe or persistent abdominal pain after intervention should be investigated using imaging findings and laboratory results to diagnose complications following thoracic duct embolization in a timely manner.

FOOTNOTES

Author contributions: Dung LV, Hien MM, Luu DT, and Linh LT prepared the case file retrieval and case summary; Dung LV, Hien MM, and Duc NM designed the research; Dung LV, Hien MM, and Tra My TT performed the research; Dung LV and Duc NM wrote the paper; all authors read and approved the final manuscript.

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