

Available online at www.sciencedirect.com

ScienceDirect

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

Case Report

Postoperative chylous ascites successfully managed by selective lymphatic embolization: Case report and literature review^{☆,☆☆}

Nguyen Ngoc Cuong, PhD^a, Nguyen Thanh Van Anh, PhD^{a,*}, Le Tuan Linh, PhD^{a,b}, Tran Quoc Hoa, PhD^c

^aDiagnostic Imaging and Interventional Center, Hanoi Medical University Hospital, No1, Ton That Tung, Dong Da, Hanoi, Vietnam

^bDepartment of Radiology, Hanoi Medical University, Hanoi, Vietnam

^cDepartment of Urology Surgery, Hanoi Medical University Hospital, No1, Ton That Tung, Dong Da, Hanoi, Vietnam

ARTICLE INFO

Article history:

Received 23 September 2024

Revised 1 November 2024

Accepted 5 November 2024

Keywords:

Postoperative chylous ascites

Thoracic duct obstruction

Embolization

ABSTRACT

Postoperative chylous ascites is a rare complication following retroperitoneal surgeries, presenting significant challenges in diagnosis and management. Retroperitoneal cyst surgery resulting in chylous leaks is an uncommon complication that has not been previously reported in the literature. Therefore, we report a clinical case of postoperative chylous ascites following retroperitoneal cyst removal with underlying idiopathic thoracic duct obstruction. This case report details the clinical features and imaging characteristics, as well as provides insights into the diagnosis of chylous leaks and the management of selective embolization of the lymphatic branches in the lumbar region.

© 2024 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Introduction

The thoracic duct is the largest lymphatic conduit, draining upwards of 75% of the body's total lymphatic flow [1]. Its anatomy is extremely complex with numerous clinical variations that can impact surgical and interventional procedures [1,2]. Spontaneous thoracic duct obstruction of unknown eti-

ology can also be considered an anatomical variation, as it may occur asymptotically in healthy individuals. However, pathological manifestations can arise under conditions such as surgery or trauma. We report a rare clinical case of chylous ascites after operation of a giant retroperitoneal cyst, in the context of idiopathic thoracic duct obstruction, which was diagnosed and managed through lymphatic embolization at Hanoi Medical University Hospital.

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

^{☆☆} Funding: Self-financed.

* Corresponding author.

E-mail address: nguyenthanhvananhnt44@gmail.com (N.T. Van Anh).

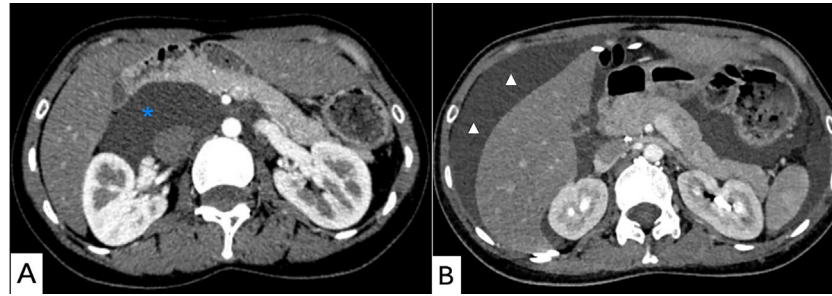


Fig. 1 – Preoperative retroperitoneal cyst and postoperative ascites. (A) Axial contrast-enhanced abdominal CT at the level of the kidneys demonstrates a large cystic structure within the retroperitoneum, measuring 103×54 mm, with a thin wall and clear fluid (blue asterisk). **(B)** Postoperative axial contrast-enhanced abdominal CT at the level of the kidneys reveals the presence of ascites (white arrowhead).

Case presentation

A 25-year-old female patient presented with abdominal distension. She had a history of surgery for a large retroperitoneal cyst (103×54 mm) 1 month prior (Fig. 1A). A few days postoperatively, the patient experienced progressive abdominal enlargement accompanied by sensations of fullness and discomfort. Clinical examination raised suspicion of ascites, which was subsequently confirmed by ultrasound and computed tomography (Fig. 1B). The aspirated fluid was milky white and had a triglyceride level greater than 100 mg/dL, confirming it as chylous fluid. The daily output of chylous fluid exceeded 1000 ml, with some days reaching 1500 ml, and persisted for 1 month. Despite conservative management with total parenteral nutrition, the chylous output remained unabated.

The patient was admitted to our hospital in a state of severe cachexia: height 165 cm, weight 40 kg (BMI 14.7). The patient was indicated for MRI lymphangiography to assess the central conducting lymphatics. This imaging technique is well-documented in the literature [3]. Under ultrasound guidance, a contrast agent was injected into the inguinal lymph nodes using a 25-gauge needle placed in each groin node. A macrocyclic gadolinium-based contrast medium (GBCM) was

used at the standard dose of 0.1 mmol/kg, diluted at a 1:1 ratio with normal saline to mitigate the T2 effects from concentrated gadolinium, which can cause signal darkening. Half of the dose was administered into each inguinal lymph node via hand injection at a slow rate (approximately 0.5–1 mL/min). MR lymphangiography was subsequently performed using a 1.5-T scanner (GE Healthcare) with a protocol including 3D FSE T2-weighted and 3D GRE T1-weighted sequences in the coronal and axial planes, with imaging captured every minute until the contrast agent reached the venous angle. MRI lymphangiography demonstrated extensive contrast extravasation into the abdominal cavity. The leakage was observed at the level of the right renal hilum, and the lower thoracic duct was visualized (Fig. 2A and B).

The patient underwent intranodal lymphangiography for selective lymphatic embolization. Under local anesthesia, a 25-gauge needle was inserted into the left inguinal lymph node under ultrasound guidance, targeting the boundary of the lymph node cortex and medulla. Subsequently, approximately 20 mL of ethiodized oil contrast medium (Lipiodol Ultra Fluid) was slowly infused into the lymph node via manual injection using a small (1–3 mL) syringe at a rate of 0.3 mL/min. Fluoroscopic and digital subtraction images were recorded at various intervals [4]. The contrast medium traversed the

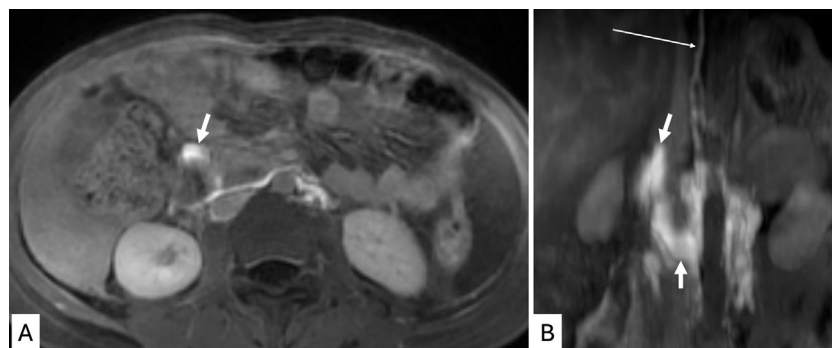


Fig. 2 – MRI lymphangiography. (A) Axial T1W fatsat sequence at the level of the kidneys shows extravasation of contrast at the level of the right renal hilum (short white arrow). **(B)** Coronal reformation of T1W sequence shows extravasation of contrast (short white arrows) with visualization of the lower thoracic duct (long white arrow).

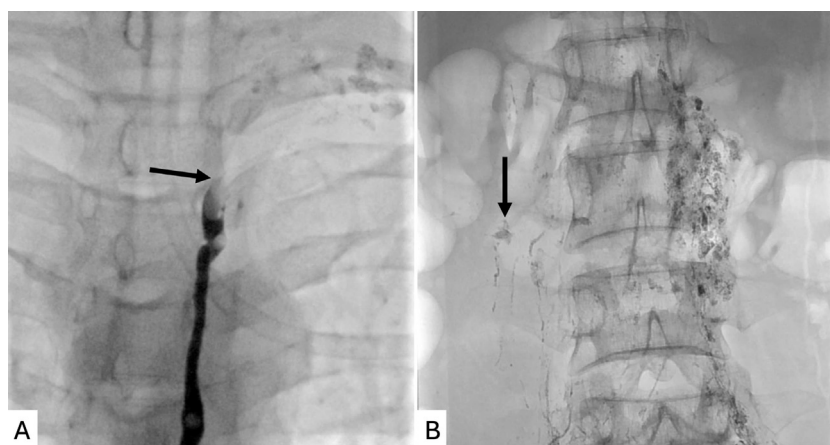


Fig. 3 – Intranodal lymphangiography. (A) Contrast material ascended from the lumbar region to the thoracic duct and then stopped at the proximal end of the thoracic duct (black arrow). (B) Extravasation of contrast is shown at the right renal hilum (black arrow).

lymphatic drainage pathways along the lymphatic vessels surrounding the bilateral iliac vessels, rising to the lumbar region. The imaging revealed that the contrast agent ascended from the lumbar region to the cisterna chyli and the thoracic duct normally; however, the contrast agent was observed to stop at the upper thoracic duct (Fig. 3A). Additionally, there was dilation of the lymphatic branches in the lumbar region at the level of both kidneys (Fig. 3B), with contrast extravasation into the abdominal cavity originating from the dilated lumbar branches.

Using a transabdominal puncture technique, a microcatheter was advanced from the cisterna chyli into the thoracic duct, where it was positioned in the lower third of the duct. This microcatheter was used to continuously inject contrast to opacify the thoracic duct. Concurrently, the opacified upper thoracic duct was punctured with a 21G Chiba needle under fluoroscopic guidance. A guidewire (Control Wire V18, Boston Scientific) was then successfully introduced retrogradely into the thoracic duct, facilitating the advancement of a second microcatheter (Tokai 1.9F) over the guidewire to the

lower thoracic duct and further into the right lumbar trunk. Contrast injection through this microcatheter revealed leakage of the contrast agent into the abdominal cavity (Fig. 4A). Cone-beam CT imaging confirmed the presence of contrast in the abdominal cavity (Fig. 4B). The diagnosis confirmed a leak from a right-sided thoracic duct branch, caused by a postsurgical injury, with an underlying condition of idiopathic thoracic duct obstruction.

The patient subsequently underwent intervention: a second microcatheter was used to administer 2.5 ml of NBCA (n-Butyl-2-cyanoacrylate) glue, diluted at a 1:3 ratio with lipiodol. During the injection of the NBCA glue into the second microcatheter, the first microcatheter was maintained in the lower thoracic duct and continuously infused with 5% glucose to prevent the glue from refluxing into the thoracic duct. On the second day postintervention, no further drainage fluid was observed. The patient transitioned to a regular diet on the third day following the procedure and was discharged on the fifth day. A follow-up examination at 6 months revealed no residual abdominal fluid.

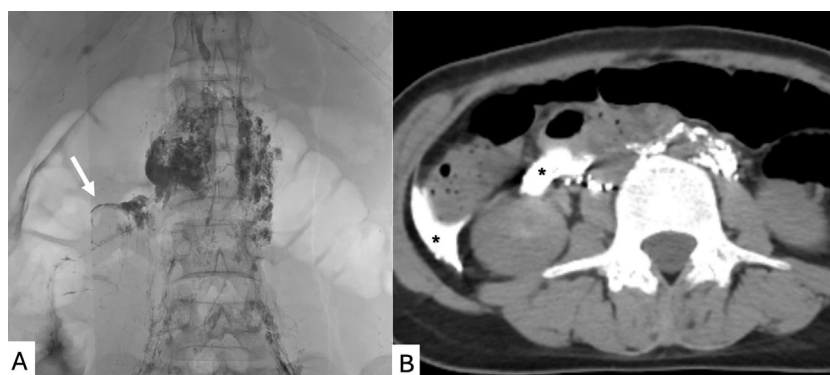


Fig. 4 – (A) The imaging from fluoroscopy confirmed extravasation of contrast from a right-sided thoracic duct branch (white arrow). (B) Cone-beam CT imaging confirmed the presence of contrast in the abdominal cavity (black asterisk).

Discussion

Congenital simple cystic lesions in the abdomen include mesenteric cysts, peritoneal simple mesothelial cysts, enteric cysts, and, more rarely, cystic lymphangioma and chylolymphatic mesenteric cysts [5]. Surgery is the treatment of choice for most abdominal cystic lesions, providing the possibility of complete excision with minimal risk of complications and recurrence [5]. Thus far, no cases of chylous leaks in the abdomen following cyst excision have been reported in the literature. Even in cases of chylous mesenteric cysts, a comprehensive review by Doreen et al. [6] involving 18 patients showed that most cysts were successfully excised, with only 2 cases reporting complications - 1 of acute pancreatitis and another of partial bowel obstruction due to adhesions - and no instances of chylous leaks were observed. Similarly, mesenteric lymphangiomas are typically completely resected, with no reported postoperative complications or recurrences [7]. In our clinical case, the patient's cystic lesion was incidentally discovered during a routine health examination, despite being asymptomatic. Although the cyst was successfully removed during surgery, the patient subsequently developed significant and persistent chylous leak, unresponsive to conservative management, ultimately leading to progressive debilitation.

Abdominal chylous leak can occur spontaneously or postoperatively. Postoperative leaks are most commonly associated with procedures involving the aorta, inferior vena cava, retroperitoneal lymphadenectomy, pancreaticoduodenectomy, gastrectomy, vagotomy, nephrectomy, and gynecological procedures, with the highest incidence observed in abdominal aortic surgeries (81%). [8–10] Leibovitch et al. [8] explains the mechanism of postoperative chylous leaks as resulting from surgical trauma to the thoracic duct, the cisterna chyli, or major lymphatic branches, coupled with increased lymphatic fluid production and obstruction of lymphatic flow in the abdominal cavity.

Similarly, Al-Busafi et al. [9] emphasizes that postoperative chylous leaks are often caused by direct trauma to the lymphatic drainage system. In the context of postoperative leaks, the studies do not mention the specific category of leaks following abdominal cyst removal. Furthermore, there are no reported cases in the medical literature of abdominal chylous leaks following the excision of a simple cyst, indicating that our clinical case is extremely rare. We hypothesize that the preoperative cyst may have been a localized lymphatic fluid or chylous collection. The surgery may have inadvertently created a connection between the lymphatic system and the abdominal cavity, leading to the development of chylous ascites, an exceptionally rare complication.

Post-MRI lymphangiography revealed: dilation of the lymphatic branches in the lumbar region at the level of both kidneys (Fig. 3B); and contrast extravasation into the abdominal cavity from the dilated lumbar branch on the right. Notably, the contrast material ascended from the lumbar region to the cisterna chyli and thoracic duct, but then stopped at the upper end of the thoracic duct, suggesting a thoracic duct obstruction despite the patient's lack of prior trauma or surgery in

the cervical region. This was confirmed during the DSA lymphangiography. Thus, it is concluded that the right branch of the thoracic duct was lacerated during the resection of the retroperitoneal cyst in the right lumbar region, on the background of thoracic duct obstruction and dilation of the lumbar lymphatic branches. Thoracic duct obstruction may result from trauma, or surgery, or occur spontaneously without an obvious cause [1,11].

Thoracic duct obstruction due to trauma or surgery typically presents with clinical symptoms of increased lymphatic system pressure, such as chylothorax or chylous ascites. In contrast, thoracic duct obstruction of unknown etiology that persists over time, with the development of collateral lymphatic pathways, may remain asymptomatic and can be present in the general population. However, due to factors such as trauma, surgical interventions, or compressive tumors that disrupt lymphatic flow, the actual pathological manifestations may become apparent [1]. This explains our clinical case: the patient exhibited obstruction of the terminal segment of the thoracic duct despite no history of cervical trauma or surgery, and previously asymptomatic presentation. However, postoperative injury to the right thoracic duct branch, occurring in the context of pre-existing thoracic duct obstruction, resulted in substantial and persistent chylous leaks into the abdominal cavity, which proved to be challenging to manage. The combination of these factors may account for the chylous leaks following abdominal cyst excision.

In terms of treatment, we selected the retrograde thoracic duct embolization technique to occlude the leaking lymphatic branches. This method has been described in various clinical reports [1,12,13]. The technique enables precise catheter placement from proximal to distal, allowing targeted embolization of the affected lumbar lymphatic branch. This targeted approach improves treatment efficacy and leads to better clinical outcomes. The patient underwent selective embolization of the leaking branch in the right lumbar thoracic duct using a mixture of NBCA: lipiodol. During the embolization, we continuously infused 5% glucose through a secondary catheter to ensure that the glue was delivered to the leak site without reflux into the thoracic duct. Postembolization results showed that the patient's chylous leaks resolved starting from the second day, with the patient being discharged on the fifth day and no recurrence after 6 months. The treatment was successful with minimal invasiveness and effective clinical outcomes.

Conclusion

Excision of a retroperitoneal cyst in the lumbar region can lead to complications such as chylous leaks into the abdomen, especially in patients with pre-existing thoracic duct obstruction and preoperative dilation of the lumbar lymphatic branches that were unknown before surgery. Selective embolization of the leaking chylous branch is an effective, minimally invasive treatment that helps prevent the need for reoperation.

Ethics approval

Our institution does not require ethical approval for reporting individual cases.

Patient consent

Informed consent for patient information to be published in this article was obtained.

REFERENCES

- [1] Johnson OW, Chick JFB, Chauhan NR, Fairchild AH, Fan CM, Stecker MS, et al. The thoracic duct: clinical importance, anatomic variation, imaging, and embolization. *Eur Radiol* 2016;26(8):2482–93.
- [2] Phang K, Bowman M, Phillips A, Windsor J. Review of thoracic duct anatomical variations and clinical implications. *Clin Anat* 2014;27(4):637–44.
- [3] Chavhan GB, Amaral JG, Temple M, Itkin M. MR lymphangiography in children: technique and potential applications. *Radiographics* 2017;37(6):1775–90.
- [4] Rajebi MR, Chaudry G, Padua HM, Dillon B, Yilmaz S, Arnold RW, et al. Intranodal lymphangiography: feasibility and preliminary experience in children. *J Vasc Interv Radiol* 2011;22(9):1300–5.
- [5] Yacoub JH, Clark JA, Paal EE, Manning MA. Approach to cystic lesions in the abdomen and pelvis, with radiologic-pathologic correlation. *Radiographics* 2021;41(5):1368–86.
- [6] Lee DLP, Madhuvrata P, Reed MW, Balasubramanian SP. Chylous mesenteric cyst: a diagnostic dilemma. *Asian J Surg* 2016;39(3):182–6.
- [7] Wani I. Mesenteric lymphangioma in adult: a case series with a review of the literature. *Dig Dis Sci* 2009;54(12):2758–62.
- [8] Leibovitch I, Mor Y, Golomb J, Ramon J. The diagnosis and management of postoperative chylous ascites. *J Urol* 2002;167(2):449–57 Pt 1.
- [9] SA Al-Busafi, Ghali P, Deschênes M, Wong P. Chylous ascites: evaluation and management. *ISRN Hepatol* 2014;2014:240473.
- [10] Combe J, Buniet JM, Douge C, Bernard Y, Camelot G. [Chylothorax and chylous ascites following surgery of an inflammatory aortic aneurysm. Case report with review of the literature]. *J Mal Vasc* 1992;17(2):151–6.
- [11] McGrath EE, Blades Z, Anderson PB. Chylothorax: aetiology, diagnosis and therapeutic options. *Respir Med* 2010;104(1):1–8.
- [12] Chen E, Itkin M. Thoracic duct embolization for chylous leaks. *Semin Intervent Radiol* 2011;28(1):63–74.
- [13] Mittleider D, Dykes TA, Cicuto KP, Amberson SM, Leusner CR. Retrograde cannulation of the thoracic duct and embolization of the cisterna chyli in the treatment of chylous ascites. *J Vasc Interv Radiol* 2008;19(2):285–90 Pt 1.