CASE REPORT



Retrograde transvenous thoracic duct embolization for lymphatic leakage after retroperitoneal tumor and lymph node resection: a case report and literature review

Go Kinoshita^{1*}, Koichi Morisaki¹, Daisuke Okamoto², Takehiko Aoyagi¹, Shinichiro Yoshino¹, Kentaro Inoue¹ and Tomoharu Yoshizumi¹

Abstract

Background Postoperative lymphatic leakage is a complication of ineffective conservative treatment for retroperitoneal mass. Herein, we report a case of lymphatic leakage that arose after retroperitoneal tumor resection and that was treated with retrograde transvenous thoracic duct embolization.

Case presentation A 28-year-old man with persistent abdominal pain was diagnosed with a large retroperitoneal metastatic tumor measuring 10 cm and a subdiaphragmatic lymph node originating from a testicular tumor. After high orchidectomy and neoadjuvant chemotherapy, the subdiaphragmatic lymph node and retroperitoneal tumor were resected together with the abdominal aorta; the latter was reconstructed using a prosthetic graft. Postoperatively, the patient developed chylothorax. No improvement was observed after conservative treatment that included fasting and somatostatin therapy. The leakage site could not be identified using antegrade lymphangiography of the bilateral inguinal lymph nodes, but was detected using retrograde transvenous lymphangiography. The leakage site was successfully embolized.

Conclusion This case report describes successful treatment with retrograde transvenous thoracic duct embolization for chylothorax following resection of a retroperitoneal tumor and lymph node. This approach is a less invasive and more effective mode of treatment for chylothorax and should be considered before surgical thoracic duct ligation when the leakage point cannot be identified using the antegrade approach.

Keywords Lymphatic leakage, Retrograde transvenous thoracic duct embolization, Chylothorax

*Correspondence:

Go Kinoshita

kinoshita.go.192@s.kyushu-u.ac.jp

¹ Department of Surgery and Science, Graduate School of Medical Sciences, Kyushu University, 3-1-1, Maidashi, Higashi-Ku,

Fukuoka 812-8582, Japan

² Department of Clinical Radiology, Graduate School of Medical Sciences, Kyushu University Hospital, 3-1-1, Maidashi, Higashi-Ku, Fukuoka 812-8582, Japan

Background

Postoperative lymphatic leakage is a relatively rare complication, occurring in 3% of postoperative esophageal cancers [1], 2–7% of retroperitoneal lymph node resections, and less than 1% of aortic aneurysm repairs [2]. Treatment for lymphatic leakage includes conservative management, somatostatin therapy, antegrade and retrograde thoracic duct embolization, percutaneous transabdominal thoracic duct embolization, or surgical thoracic duct ligation [3]. The first choice is conservative treatment with fasting and somatostatin therapy placement of



© The Author(s) 2024. **Open Access** This article is licensed under a Creative Commons Attribution 4.0 International License, which permits use, sharing, adaptation, distribution and reproduction in any medium or format, as long as you give appropriate credit to the original author(s) and the source, provide a link to the Creative Commons licence, and indicate if changes were made. The images or other third party material in this article are included in the article's Creative Commons licence, unless indicated otherwise in a credit line to the material. If material is not included in the article's Creative Commons licence and your intended use is not permitted by statutory regulation or exceeds the permitted use, you will need to obtain permission directly from the copyright holder. To view a copy of this licence, visit http://creativecommons.org/licenses/by/4.0/.

a drainage duct. However, when the volume of lymphatic fluid exceeds 1000 mL/day, conservative treatment alone is unlikely to cure the leakage [4]. If conservative treatment is unsuccessful, invasive treatment may be administered that includes lymphangiography with thoracic duct embolization, transabdominal embolization, and surgical thoracic duct ligation [1, 5, 6]. With respect to lymphangiography, two approaches have been reported: the antegrade approach from the inguinal lymph nodes and the retrograde transvenous approach. An antegrade approach is usually selected, but its success rate is only 70% [7]. The retrograde transvenous approach was first reported by Mittleider et al. in 2008 [1]. Recently, Jun et al. reported that the retrograde approach combined with the antegrade approach may improve the treatment outcome of thoracic duct embolization [7]. We report a case of chylothorax after retroperitoneal tumor resection treated using the antegrade approach via the inguinal lymph node and retrograde transvenous thoracic duct embolization.

Case presentation

A 28-year-old man with persistent abdominal pain was examined and found to have a large retroperitoneal tumor measuring 10 cm diameter and a testicular tumor. A retroperitoneal tumor involving the superior mesenteric artery, bilateral renal arteries, and abdominal aorta, and an enlarged subdiaphragmatic lymph node were revealed on computed tomography (Fig. 1a-c). In accordance with the guidelines for the treatment of testicular tumors, the patient underwent high orchidectomy. Histological examination showed that the tumor was a seminoma. After chemotherapy-four courses of bleomycin, etoposide, and platinum therapy; two courses of etoposide and cisplatin therapy; and two courses of paclitaxel, ifosfamide, and cisplatin therapy—and shrinkage of the tumor (Fig. 1d, e), the subdiaphragmatic lymph node and retroperitoneal tumor with the abdominal aorta were resected. The abdominal aorta was resected from infrarenal aorta to terminal aorta and reconstructed with a prosthetic graft (Fig. 1f, g). On the fourth day after surgery, a chest radiograph showed massive pleural effusion (Fig. 2a) and a drainage tube was inserted into the patient's right chest cavity (Fig. 2b). When the patient resumed food intake on the seventh day after surgery, the pleural effusion changed from serous to chyle, and the patient was diagnosed with lymphatic leakage. The patient was administered conservative treatment consisting of fasting and somatostatin therapy (300 μ g/



Fig. 1 Computed tomography (CT) images and intraoperative findings. **a** The three-dimensional CT image captured before chemotherapy. The dotted line delineates the extent of the tumor. The black arrow indicates the ureteral stent. **b**, **c** CT images before chemotherapy. **d**, **e** CT images after chemotherapy. **f** Intraoperative findings. The dotted line indicates the extent of the tumor. **g** Intraoperative findings after tumor resection with abdominal aorta and reconstruction by prosthetic graft



Fig. 2 Chest radiograph images. **a** Chest radiograph captured on postoperative day 4. Large pleural effusion is visible within the area delineated by the dotted line. **b** After thoracic drain placement; the black arrow indicates the drain

day); however, the volume of fluid drained continued at approximately 1000 mL/day.

On the thirteenth postoperative day, antegrade lymphangiography of the bilateral inguinal lymph nodes was performed; however, no lymphatic leakage was identified (Fig. 3). On the fifteenth day after surgery, a lymphangiography using a retrograde transvenous approach was performed. The procedure of retrograde transvenous approach was shown in the schema (Fig. 4). This schema was created based on the report by Sato et al. [8]. The left cephalic vein was percutaneously punctured and an 11-cm 7 Fr sheath was placed. A 7 Fr 65-cm KMP catheter was advanced to the venous angle using a 0.035-in. 180-cm Radifocus guidewire (Terumo, Tokyo, Japan) (Fig. 5a). Cannulation of the thoracic duct was performed using a 0.014-in. 180-cm Asahi Chikai V guidewire (ASAHI INTECC, Aichi, Japan) supported by a 2.0/2.4 Fr 150cm Leonis Mova catheter (SB-KAWASUMI LABO-RATORIES, Kanagawa, Japan) (Fig. 5b). The catheter was advanced to the eleventh thoracic vertebra level through several thoracic duct valves. Lymphangiography revealed interruption of the thoracic duct and spillage of contrast with Lipiodol outside the duct (Fig. 5c). The thoracic duct was embolized with 0.4 mL of a 1:2 mixture of n-butyl-2-cyanoacrylate and Lipiodol. After thoracic duct embolization, the drainage volume decreased to 100 mL/day, and the drainage tube was removed. The patient was discharged on the twenty-ninth day after surgery without recurrent effusions.



Fig. 3 Contrast image after the antegrade lymphangiography. The circles show the reach of Lipiodol contrast. Lipiodol contrast from the right inguinal lymph nodes flowed into the vein. Lipiodol contrast from the left inguinal lymph nodes reached near the left kidney



Fig. 4 Schema of the retrograde transvenous approach. 1. Puncture the arm vein and place a sheath. 2. Advance the guidewire and catheter and cannulate the entrance of the thoracic duct. 3. Advance the guidewire and catheter into the thoracic duct



Fig. 5 Contrast images. a Black arrows indicate the left internal jugular and subclavian veins. The angle created by the two veins is the venous angle. b The wire and catheter are advancing into the thoracic duct. c Contrast image of the thoracic duct. The thoracic duct is disrupted at the dotted line

Discussion

In the present case, the postoperative chylothorax was successfully treated with retrograde transvenous thoracic duct embolization after the antegrade approach failed. The failure of the antegrade approach in this case may have occurred because the distal side of the thoracic duct was tightly ligated during resection of the subdiaphragmatic lymph node. As demonstrated in this case, the retrograde transvenous approach is an effective and less invasive treatment than the percutaneous transthoracic or abdominal approach, or surgical thoracic duct ligation.

With respect to the transthoracic or abdominal approaches, Itkin et al. reported a success rate for embolization of 68.9% (73/106 cases), a clinical success rate of 81.7% (58/71 cases), and a complication rate of 2.8% (3/71 cases) [3]. Pamarthi et al. reported a success rate

Authors	Year	Number of patients	Age	Sex	Causes of lymph leakage	Technical success	Embolization success	Clinical success	Complications
Mittleider et al. [1]	2008	1	60 s	M:1	Postoperative pan- creatic cancer:1	1/1	1/1	0/1	None
Koike et al. [9]	2013	2	50-60 s	M:1, F:1	Postoperative thoracic aortic aneurysm:1 spinal arteriovenous malfor- mation:1	2/2	2/2	1/2	None
Schild et al. [10]	2015	1	-	-	-	1/1	1/1	1/1	None
Sugimura et al. [6]	2017	1	60 s	M:1	Liver cirrhosis:1	1/1	1/1	1/1	None
Arslan et al. [11]	2017	1	50 s	F:1	Cervical lymph node dissection:1	1/1	1/1	1/1	None
Hussain et al. [12]	2018	1	20 s	M:1	Traffic trauma:1	1/1	1/1	1/1	None
Srinivasa et al. [2]	2018	3	10-70 s	M:2, F:1	Post-nephrectomy:2, hypoplastic left heart:1	3/3	3/3	3/3	None
Majdalany et al. [13]	2018	3	60-70 s	M:2, F:1	Post- nephrectomy:2,post- pancreatectomy and splenectomy	3/3	3/3	2/3	None
Kariya et al. [14]	2018	13	20-80 s	M:5, F:8	Lymphangiectasis:3, thyroid cancer:1, systemic lupus erythematosus:1, acquired immunode- ficiency syndrome:1, past esophageal cancer chemoradia- tion:1, postoperative esophageal cancer:1, gastric cancer sur- gery:2, lung cancer:1, cervical cancer:1, unknown:1	8/13	1/1	1/1	Thoracic valve injury
Rott et al. [15]	2020	1	60 s	F:1	Postoperative Nissen surgery:1	1/1	1/1	1/1	None
Morikawa et al. [5]	2020	3	40-70 s	M:1, F:2	Postoperative par- aaortic lymph node dissection, post- nephrectomy:2	3/3	3/3	3/3	None
Dar et al. [16]	2021	1	50 s	M:1	Traffic trauma:1	1/1	1/1	1/1	None
Sato et al. [8]	2021	1	70 s	M:1	Postoperative esophageal cancer:1	1/1	1/1	1/1	None
Kalia et al. [17]	2022	2	50 s	F:2	Post-nephrectomy:2	1/1	1/1	1/1	None
Seth et al. [18]	2022	1	50 s	M:1	Unknown:1	1/1	1/1	1/1	None
Jun et al. [7]	2022	6	-	_	-	-	-	-	-
Petrini et al. [19]	2023	1	60 s	M:1	Non-Hodgkin lym- phoma:1	1/1	1/1	1/1	None
Kim et al. [20]	2023	16	-	-	-	8/16	8/16	6/12	-

Table 1 Cases in literature reporting retrograde transvenous thoracic duct embolization

for embolization of 79.2% (95/120 cases), a clinical success rate of 50.8% (61/120 cases), and a complication rate of 5.8% (7/120 cases) [4]. The most common complication associated with this procedure was influx of embolic material into the pulmonary artery, which did not require

additional treatment [3, 4]. In cases where lymph nodes have been extensively resected, the problem that arises is that transabdominal puncture cannot be performed because the flow of lymph fluid is obstructed and the site of the thoracic duct injury cannot be identified using lymphangiography [5]. Furthermore, the transabdominal approach carries the risk of organ penetration, including the arteries, biliary system, and intestines.

Antegrade thoracic duct embolization is performed by puncturing the inguinal lymph node under ultrasound guidance [3]. The technical success rate of the antegrade approach is approximately 70% [3, 4, 7]. As in the present case, Jun et al. showed that using a retrograde transvenous approach after failure of the antegrade approach could improve the technical success [7]. The retrograde transvenous approach was first reported by Mittleider et al. in 2008 [1].

We conducted a review of studies in which the retrograde transvenous approach was used and a summary of the findings is presented in Table 1. We reviewed the literature published between 2008 and September 2023, and retrieved the details of only 58 cases reported in English by 18 authors. In the reports retrieved, the success rates of cannulation and lymphatic leakage embolization were 73.6% (39/53 cases) and 80% (32/40 cases), respectively. The clinical success rate was 75.0% (27/36 cases) and the complication rate was 2.8% (1/36 cases).

The retrograde transvenous approach is a less invasive and more effective treatment for chylothorax; however, it may be considered technically more challenging. The end of the thoracic duct has an ostial valve, which is usually difficult to visualize, and the confluence of the subclavian vein and the thoracic duct is difficult to cannulate because of the variety of veins. Furthermore, valves in the thoracic duct and variations in the thoracic duct may prevent the contrast agent from reaching the injured area. When a retrograde venous approach is used, lymphangiography from the inguinal lymph nodes should be used to identify the entry site of the thoracic duct [5]. If lymphangiography from the inguinal lymph node is not available, lymphangiography should be performed beforehand using magnetic resonance imaging to determine the location around the thoracic duct and the vein. Balloon-occluded retrograde lymphangiography may also be useful [12]. A 5 Fr balloon catheter is inserted into the thoracic duct, and a contrast agent is released from the tip of the balloon catheter with balloon inflation for contrast. By blocking lymphatic drainage, the lower thoracic duct may be contrasted and the site of injury identified.

Another factor that complicates the retrograde transvenous approach is the presence of several patterns in which the thoracic duct may enter the venous angle. The cervical part of the thoracic duct is defined as the distance from the beginning of the thoracic duct to the level of the left brachiocephalic vein, whereas the thoracic part is defined as the remaining section of the thoracic duct [14]. At the cervical part, the plexiform type is defined as having a plexiform configuration without a prominent main duct. In the case of the plexiform type, advancing the catheter to the thoracic duct is difficult, although cannulation to the thoracic entrance was possible in all three cases [14]. Thus, the retrograde transvenous approach is difficult to perform in cases where the cervical part is of the plexiform type.

Conclusions

This case report describes successful treatment with retrograde transvenous thoracic duct embolization for chylothorax following the resection of a retroperitoneal tumor and lymph node. This approach is a less invasive and more effective treatment for chylothorax and should be considered before surgical thoracic duct ligation when the leakage site cannot be identified using an antegrade approach.

Abbreviation

CT Computed tomography

Acknowledgements

We would like to thank editage for English language editing.

Author contributions

GK and KM collected, analyzed, and interpreted the data and revised the manuscript. All authors read and approved the final manuscript.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Availability of data and materials

Not applicable.

Declarations

Ethics approval and consent to participate Not applicable.

Consent for publication

Informed consent was obtained from the patient for the publication of this report.

Competing interests

All authors declare that there is no conflict of interest.

Received: 31 December 2023 Accepted: 4 March 2024 Published online: 08 March 2024

References

- 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;19:285–90.
- Srinivasa RN, Gemmete JJ, Osher ML, Hage AN, Chick JFB. Endolymphatic balloon-occluded retrograde abdominal lymphangiography (BORAL) and embolization (BORALE) for the diagnosis and treatment of chylous

ascites: approach, technical success, and clinical outcomes. Ann Vasc Surg. 2018;49:49–56.

- Itkin M, Kucharczuk JC, Kwak A, Trerotola SO, Kaiser LR. Nonoperative thoracic duct embolization for traumatic thoracic duct leak: experience in 109 patients. J Thorac Cardiovasc Surg. 2010;139:584–90.
- Pamarthi V, Stecker MS, Schenker MP, Baum RA, Killoran TP, Suzuki Han A, et al. Thoracic duct embolization and disruption for treatment of chylous effusions: experience with 105 patients. J Vasc Interv Radiol. 2014;25:1398–404.
- Morikawa K, Takenaga S, Hasumi J, Kano A, Tatsuno S, Michimoto K, et al. Retrograde transvenous lymphatic embolization for postoperative chylous ascites: a report of three cases and literature review. Radiol Case Rep. 2020;15:1623–8.
- Sugimura H, Cong Y, Yamazaki I, Nomori H, Ito Y, Takeshi A. A case of nontraumatic chylothorax treated by percutaneous transvenous embolization of the thoracic duct. J Jpn Assoc Chest Surg. 2017;31:122–6.
- Jun H, Hur S, Jeong YS, Kang CH, Lee H. Thoracic duct embolization in treating postoperative chylothorax: does bail-out retrograde access improve outcomes? Eur Radiol. 2022;32:377–83.
- Sato Y, Tanaka Y, Imai T, Kawada H, Okumura N, Matsuhashi N, et al. Chylothorax after esophagectomy treated with inguinal intranodal lymphangiography and transvenous retrograde thoracic duct embolization. Clin J Gastroenterol. 2021;14:969–74.
- Koike Y, Nishimura JI, Hirai C, Moriya N, Katsumata Y. Percutaneous transvenous embolization of the thoracic duct in the treatment of chylothorax in two patients. J Vasc Interv Radiol. 2013;24:135–7.
- Schild HH, Naehle CP, Wilhelm KE, Kuhl CK, Thomas D, Meyer C, et al. Lymphatic interventions for treatment of chylothorax. Rofo. 2015;187:584–8.
- Arslan B, Masrani A, Tasse JC, Stenson K, Cenk TÜ. Superselective retrograde lymphatic duct embolization for management of postoperative lymphatic leak. Diagn Interv Radiol. 2017;23:379–80.
- Hussain JS, Srinivasa RN, Srinivasa RN, Patel A, Gemmete JJ, Chick JFB. Balloon-occluded retrograde lymphangiography and embolization of a posttraumatic lymphoenteric fistula. J Vasc Interv Radiol. 2018;29:1032–3.
- Majdalany BS, Khayat M, Downing T, Killoran TP, El-Haddad G, Khaja MS, et al. Lymphatic interventions for isolated, iatrogenic chylous ascites: a multi-institution experience. Eur J Radiol. 2018;109:41–7.
- Kariya S, Nakatani M, Ueno Y, Yoshida A, Ono Y, Maruyama T, et al. Transvenous retrograde thoracic ductography: initial experience with 13 consecutive cases. Cardiovasc Intervent Radiol. 2018;41:406–14.
- Rott G, Boecker F. High-output chyloperitoneum following laparoscopic Nissen fundoplication treated with retrograde transvenous thoracic duct embolization. CVIR Endovasc. 2020;3.
- Dar PMUD, Gamanagatti S, Priyadarshini P, Kumar S. Traumatic chylothorax: a dilemma to surgeons and interventionists. BMJ Case Rep. 2021;14:e238961.
- Kalia S, Narkhede A, Yadav AK, Bhalla AK, Gupta A. Retrograde transvenous selective lymphatic duct embolization in post donor nephrectomy chylous ascites. CEN Case Rep. 2022;11:1–5.
- Seth R, Gupta A, Yadav A. Lymphangiography and thoracic duct embolization (both antegrade and retrograde approaches) in a case of spontaneous chylous leak after failed thoracic duct ligation. J Vasc Surg Cases Innov Tech. 2022;8:779–80.
- Petrini M, Colombi D, Bodini FC, Morelli N, Ciatti C, Maniscalco P, et al. Thoracic duct leakage in a patient with type B-non-Hodgkin lymphoma treated with transvenous retrograde access embolization: a case report. Acta Biomed. 2023;94(S1):1–5.
- Kim H, Hyun D, Shin SW, Jeong G, Kim J, Cho JH, et al. Factors contributing to successful transvenous retrograde thoracic duct cannulation. J Vasc Interv Radiol. 2023;34:205–11.

Publisher's Note

Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.