

Interventional Treatment of Lymphatic Leakage Post Appendectomy: Case Report

Nguyen Ngoc Cuong¹, Nguyen Thai Binh¹, Phan Nhan Hien¹, Nguyen Hoang², Le Tuan Linh¹, Doan Tien Luu¹, Vu Dang Luu³, Pham Duc Huan², Vo Truong Nhu Ngoc⁴, Md Jamal Uddin⁵, Vu Thi Nga⁶, Dinh-Toi Chu⁷

¹Radiology Department, Hanoi Medical University Hospital, Hanoi, Vietnam; ²Department of Surgery B, Hanoi Medical University Hospital, Hanoi, Vietnam; ³Radiology Department, Bach Mai Hospital, Hanoi, Vietnam; ⁴School of Odontostomatology, Hanoi Medical University, Hanoi, Vietnam; ⁵College of Pharmacy, Ewha Womans University, Seoul, South Korea; ⁶Institute for Research and Development, Duy Tan University, Danang, Vietnam; ⁷Faculty of Biology, Hanoi National University of Education, Hanoi, Vietnam

Abstract

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***Correspondence:** Dinh Toi Chu, Faculty of Biology, Hanoi National University of Education, Hanoi, Vietnam. E-mail chudinhtoi.hnue@gmail.com

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BACKGROUND: Postoperative lymphatic complications are not common, and lymphatic leakage complication post appendectomy (LLCPC) is even rarer. However, the number of this operation is high so LLCPC can occur.

CASE REPORT: Here, we report a female patient post appendectomy with severe chylous ascites. This patient underwent six operations. A leakage point at the right iliac-fossa, which was embolized successfully after two sessions, was spotted during intranodal lymphangiography. After 6 months, the ascites were significantly reduced while some lymphatic aneurysms still existed in the lumbar-retroperitoneal region.

CONCLUSIONS: Basing the knowledge of this clinical case and literature, we have concluded that lymphatic leakage can be diagnosed and embolized by percutaneous intervention.

Introduction

Postoperative lymphatic complications include the phenomena according to a recent review: lymphocele, lymph ascites, lymphorrhea, lymphatic fistula, chylous ascites, chylothorax, chyloretroperitoneum and chyloorrhea [1]. Most lymphatic leakages can be spontaneously healed without the need for intervention; the priority treatment is conservative treatment including drainage, parenteral nutrition along with the use of somatostatin [1], [2]. However, to some death threatening cases,

prompt treatments are required [2], [3]. Various causes have been recognized as the obstacles of operated obliteration of lymphatic leakage: 1) lymphatic vessels seem to be “invisible” on macroscopic anatomy in surgery; 2) high anatomic variations between patients; 3) lymphatic injury has various types without a common treatment for each type; 4) understandings about lymphatic anatomy has been still limited [2], [3], [4].

The improvement of ultrasound-guided intranodal lymphangiography has disclosed various forms of lymphatic injury, which is favourable for the strategies of treatment [4]. Recently, the percutaneous

intervention has been applied to postoperative lymphatic injury with the support of imaging modalities including fluoroscopy and computerised tomography (CT) [2], [4], [5], [6].

Here, we report a rare case of a patient with lymphatic leakage after appendectomy.

Case Report

A thirty-three-year-old Vietnamese female with normal medical history after appendectomy due to appendicitis had her abdomen swollen quickly. Ultrasound examination revealed that she had a large amount of abdominal fluid and right pleural effusion. The colour and biochemical results of fluid sampling by fine needle aspiration confirmed that she had chylous ascites and right chylothorax. She was then followed by six different surgeries including thoracic open and abdomen surgical opening to ligate the lymphatic vessels and the thoracic duct, but all approaches were failures.

The patient came to our hospital with abdominal distention and physical exhaustion. Ultrasonography and computed tomography revealed a large amount of free abdominal fluid and right pleural cavity (Figure 1A). About 5 litres of ascites (Pigtail 6Fr- Biotech) drained out as milky white fluid (Figure 1B). Biochemical tests of the drainage fluid showed a very high concentration of triglyceride (7.1 mmol/l).

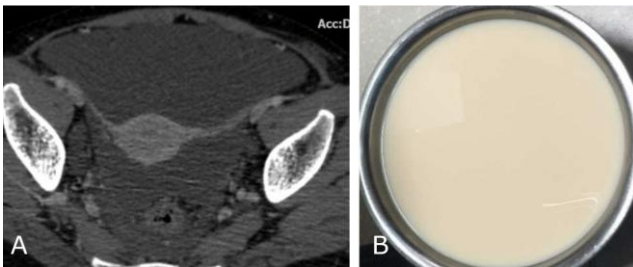


Figure 1: Abdominal CT scanner and fluid before treatment; Abdominal CT scanner revealed a large amount of fluid intraperitoneal and right pleural cavities (not shown) (A); Fluid obtained by paracentesis from ascites in milky white (B)

Lymphangiography was performed using an intranodal technique using system DSA with contrast injection to lymph nodes at both inguinal regions. The lymph node at both groins was punctured under sonography guidance with the 25-gauge needle. Contrast material (lipiodol) which was heated (up to 37°C) to reduce viscosity was injected with a recommended volume of 0.2- 0.4 ml per minute. When lymphatic vessels were opacified, we found lymphatic lesion at the right iliac fossa: a lymphatic vessel at the right iliac fossa enlarged like multi fusiform lymphatic aneurysms. From the biggest

aneurysm, there was an extravasation of lymph directly into the peritoneal cavity (Figure 2). Our diagnosis was that the patient had multi fusiform lymphatic aneurysms and there was a rupture of the biggest aneurysm into the abdomen cavity. The lymphatic aneurysms may result from the reflux of lymph within the vessel valves' pathology.

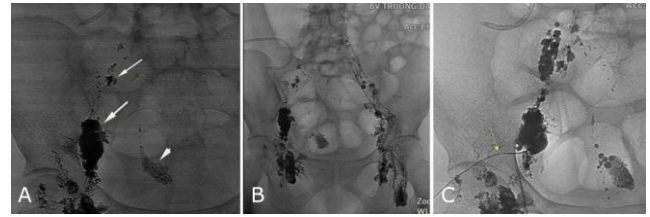


Figure 2: Lymphatic aneurysm and lymphatic extravasation; A lymphatic vessel is enlarged to the size of multi fusiform aneurysm (long arrow) and the presence of extravasation contrast in the peritoneal cavity (arrowhead) (A); Normal pattern of lymphatic channels at the left area (B); A 22-gauge-needle was punctured under guidance of DSA to perform embolization (arrow) (C)

To occlude the ruptured aneurysm, a needle (Chiba 22 G, Cook) was percutaneously punctured under the guidance of fluoroscopy into the lymphatic aneurysm. We then injected 5 ml contrast (Xenetic 350) through the needle to affirm that the tip of the needle was placed in the aneurysm. A guide wire 0.014" (transcend 14, Boston Scientific) was inserted into the lymphatic aneurysm through the needle. Through the guide wire, the 4-French-sheath was cannulated. One coil (5/20 mm, Axium 3D, EV3) was pushed into the aneurysm through the sheath. After that, we injected 10 ml mixture of n-butyl cyanoacrylate (NBCA) which was diluted with lipiodol at the ratio 1:2. After the procedure, the drainage still came out about 1 litre per 24 hours during 2 days. Abdominal CT scanner showed that the lymphatic aneurysm was partially filled with an embolic agent; where remained some compartments that were not filled with embolic agents (Figure 3). For those reasons, we decided to do the second intervention.

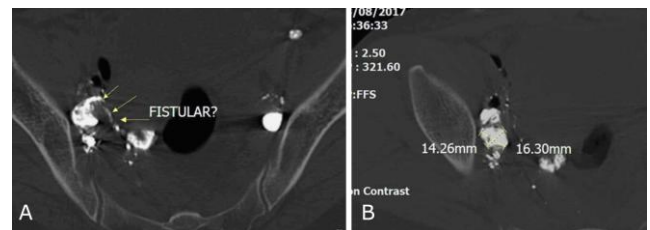


Figure 3: Abdominal CT scanner one day after the first intervention; Embolic agents filled in the aneurysm and the fistula (arrow) (A); In another hand, there might be some compartments in the aneurysm which had not been filled by embolic agent, resulting in existing leakage (B)

The second procedure was performed after 2 days. Because the aneurysm was still opacified, we punctured it with a 22-G needle under fluoroscopy. When the needle tip was in the aneurysm, we injected the sclerosis agent with the expectation that the

inflammation reaction within the aneurysm can collapse all the compartments and heal the fistular. The sclerosant used was lauromacrogol 400 (Aetoxisclerol 2%-KreusslerPharma, France) (10ml mixture of foam solution containing 2ml of laucromacrogol and 8 ml of air). Finally, we embolized the lymph node by a technique that was described as “closet upstream lymph node embolisation” in the literature [2]; 1 ml of NBCA (history/lipiodol 1:6 ratio) was injected into the lymph node at the right groin. After the second intervention, the amount of fluid leakage decreased significantly (Figure 4).

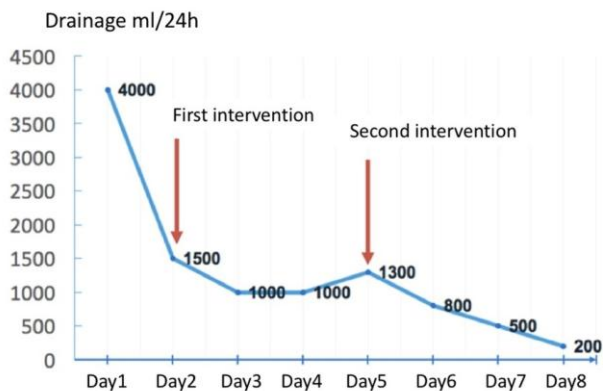


Figure 4: The amount of fluid drainage decreased after the first and second intervention

The patient was discharged one week later when the drain was empty. After six months, computed tomography revealed no more abdominal fluid, but there were still some retroperitoneal lymphatic aneurysms along the right iliac vessels that were not indicated for interventions, the patient was still under supervision (Figure 5).

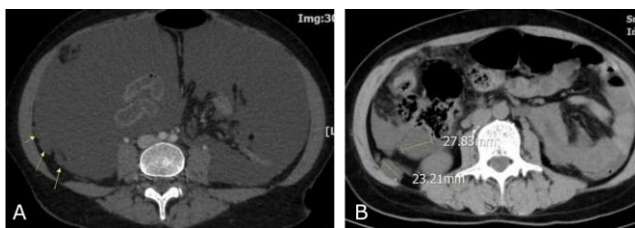


Figure 5: CT scanner of the patient before (A) and after treatment (B) showed a significant decrease of abdominal fluid; Some lymphatic aneurysms (arrow) before and after the intervention still endure, but there is no complication recognised, no further interventions have been indicated

Discussion

A lymphatic aneurysm is defined as a focal dilatation of lymphatic vessels with inflow and outflow to the normal lymphatic vessels (Figure 2A). It can appear during a long time of reflux of the lymph into

the peripheral lymphatic vessels. Percutaneous injection of the ruptured aneurysm was performed with a 22-gauge Chiba needle. The needle was punctured into the aneurysm revealed by lymphatic fluid coming out of the needle. The mixture ratio of NBCA and lipiodol was about 1:1 or 1:2 that was injected in the aneurysm. The injection time was about 10-30 seconds before the removal of the entire system. In our patients, we noticed that the leakage was high flow when injecting the contrast, so we decide to put coil before NBCA to prevent the unexpected migration of glue.

The fibrous agent used was lauromacrogol 400 (Aetoxisclerol, France) which destroyed the inner layer of the aneurysm or lymphatic vessels. Some studies have shown that sclerotherapy had a success rate of 77-90% for patients with lymphocele [6], [7]. It is necessary for the sclerosant therapy to be combined with intravenous nutrition after 2-3 days to reduce volume of lymph in the lymphatic circulation and induce the effect of sclerosant. Some authors suggest to use somatostatin along with intravenous nutrition [1], [8]. Somatostatin is a peptide hormone that is highly effective in patients with lymphatic leakage. The mechanism is supposed that it may reduce lymphatic production and concentrate the lymphatic liquid [8].

The closest lymph node is the lymph node from which efferent extravasated vessels on lymphangiography (Figure 6A). A 26-gauge needle was punctured into the lymph node under ultrasound guidance to inject the embolic liquid NBCA. Depending on the flow velocity in the *lymphatic vessels*, the NBCA to lipiodol ratio was adjusted appropriately, usually from 1:3 to 1:9, according to the experiences of operator. The injection lasted for 10 - 30 seconds so that the mixture filled the lymph node and entered lymphatic vessels [6], [9]. In our patient, when injecting contrast media into the lymph node, we noticed the flow of lymph was slow. Therefore, we chose the ratio NBCA/lipiodol at 1:2.

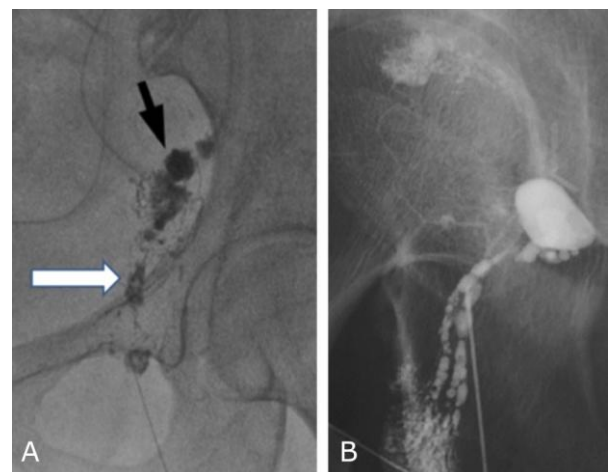


Figure 6: Lymph nodes closet to the lesion (white arrow) with pseudo aneurysm (black arrow); The needle is punctured into a lymph node at the pelvic area (A) [12]; 21G Needle is punctured into lymphatic channels just under the pseudoaneurysm (B)

The upstream lymphatic vessel is the vessel that goes directly into the lymphatic lesion on lymphangiography. It can be directly punctured with a 21-gauge needle [5]. Embolisation of this vessel can be done through the needle or a catheter by using glue NBCA. The ratio of historic/lipiodol was about 1:1.5 [2] (Figure 6B). Percutaneous intervention may have potential risks including infection and bleeding. However, no damage has been recorded even with the usage of the large needle like the 21 gauge [10], [11]. The most severe complication that may occur in lymphangiography is pulmonary artery occlusion due to embolic lipiodol flowing to the thoracic duct and then to the right ventricular. To prevent the risk, lipiodol volume should be limited under 10 ml [2], [5], [6]. Contraindications of the lymphangiography include the patients with respiratory insufficiency and left-to-right cardiac shunt.

In conclusion, intranodal lymphangiography with lipiodol is a revolutionary advance in the diagnosis of the lymphatic leakage. In our patient, this method is safe and effective. Also, percutaneous intervention to embolize the leakage with different embolic agents can become the first-line option as a less invasive, but highly effective method, which could replace the surgical treatment for patients.

Ethical Approval

All procedures performed in studies involving human participants were by the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This study was approved by the institutional review board of Hanoi Medical University Hospital

Informed Consent

Informed consent was obtained from the patient included in the study

References

- Shulan LV et al. Review postoperative lymphatic leak. *Oncotarget*. 2017; 8(40):69062-69075. <https://doi.org/10.18632/oncotarget.17297>
- Hur S, Shin JH, Lee IJ, Min SK, Min SI, Ahn S, Kim J, Kim SY, Kim M, Lee M, Kim HC. Early experience in the management of postoperative lymphatic leakage using lipiodol lymphangiography and adjunctive glue embolisation. *Journal of Vascular and Interventional Radiology*. 2016; 27(8):1177-86. <https://doi.org/10.1016/j.jvir.2016.05.011> PMID:27373491
- Yoshimatsu R, Yamagami T, Miura H, Matsumoto T. Prediction of therapeutic effectiveness according to CT findings after therapeutic lymphangiography for lymphatic leakage. *Japanese journal of radiology*. 2013; 31(12):797-802. <https://doi.org/10.1007/s11604-013-0252-2> PMID:24158649
- Lv S, Wang Q, Zhao W, Han L, Wang Q, Batchu N, Ulain Q, Zou J, Sun C, Du J, Song Q. A review of the postoperative lymphatic leakage. *Oncotarget*. 2017; 8(40):69062-69075. <https://doi.org/10.18632/oncotarget.17297> PMID:28978181 PMCid:PMC5620321
- Baek Y, Won JH, Chang SJ, Ryu HS, Song SY, Yim B, Kim J. Lymphatic embolization for the treatment of pelvic lymphoceles: preliminary experience in five patients. *Journal of Vascular and Interventional Radiology*. 2016; 27(8):1170-6. <https://doi.org/10.1016/j.jvir.2016.04.011> PMID:27241389
- Lee EW, Shin JH, Ko HK, Park J, Kim SH, Sung KB. Lymphangiography to treat postoperative lymphatic leakage: a technical review. *Korean journal of radiology*. 2014; 15(6):724-32. <https://doi.org/10.3348/kjr.2014.15.6.724> PMID:25469083 PMCid:PMC4248627
- Mahrer A, Ramchandani P, Trerotola SO, Shlansky-Goldberg RD, Itkin M. Sclerotherapy in the management of postoperative lymphocele. *Journal of Vascular and Interventional Radiology*. 2010; 21(7):1050-3. <https://doi.org/10.1016/j.jvir.2010.03.014> PMID:20537556
- Kim EA, Park H, Jeong SG, Lee C, Lee JM, Park CT. Octreotide therapy for the management of refractory chylous ascites after a staging operation for endometrial adenocarcinoma. *Journal of Obstetrics and Gynaecology Research*. 2014; 40(2):622-6. <https://doi.org/10.1111/jog.12183> PMID:24118223
- Inoue M, Nakatsuka S, Yashiro H, Tamura M, Suyama Y, Tsukada J, Ito N, Oguro S, Jinzaki M. Lymphatic intervention for various types of lymphorrhea: access and treatment. *Radiographics*. 2016; 36(7):2199-211. <https://doi.org/10.1148/rg.2016160053> PMID:27831840
- Itkin M, Kucharczuk JC, Kwak A, Trerotola SO, Kaiser LR. Nonoperative thoracic duct embolization for traumatic thoracic duct leak: experience in 109 patients. *The Journal of thoracic and cardiovascular surgery*. 2010; 139(3):584-90. <https://doi.org/10.1016/j.jtcvs.2009.11.025> PMID:20042200
- Nadolski GJ, Itkin M. Thoracic duct embolization for nontraumatic chylous effusion: experience in 34 patients. *Chest*. 2013; 143(1):158-63. <https://doi.org/10.1378/chest.12-0526> PMID:22797603
- Gemmete JJ, Srinivasa RN, Chick JF. Treatment of Chylous Ascites in a Child after Wilms Tumor Resection with Intranodal Injection of N-Butyl Cyanoacrylate Glue. *Journal of vascular and interventional radiology: JVIR*. 2017; 28(7):1067-1069. <https://doi.org/10.1016/j.jvir.2017.02.024> PMID:28645505