Ultrasound-guided intranodal lipiodol lymphangiography from the groin is useful for assessment and treatment of post-esophagectomy chylothorax in three cases

Jiajia Liu a, Yusuke Sato a,⁎, Satoru Motoyama a, Koichi Ishiyama b, Kentaro Yamada c, Masayoshi Yamamoto c, Akiyuki Wakita a, Yuta Kawakita a, Kazuhiro Imai a, Hajime Saito a, Manabu Hashimoto b, Yoshihiro Minamiya a

a Department of Thoracic Surgery, Akita University Graduate School of Medicine, Akita, Japan
b Department of Radiology, Akita University Graduate School of Medicine, Akita, Japan
c Department of Radiology, National Defense Medical College, Tokorozawa, Japan

ARTICLE INFO

Article history:
Received 13 October 2016
Accepted 30 October 2016
Available online 3 November 2016

Keywords:
Intranodal lymphangiography
Lipiodol
Postoperative chylothorax

ABSTRACT

INTRODUCTION: Ultrasound-guided intranodal lipiodol lymphangiography (LAG) from the groin is a recently introduced technique for diagnosing and treating postoperative chylothorax. The benefits of this technique include reduced technical difficulty and shorter procedure duration, as compared to traditional post-esophagectomy LAG. Although these benefits may eventually increase utilization of intranodal LAG, reports are still few.

PRESENTATION OF CASES: Herein, we report three cases of post-esophagectomy chylothorax in whom ultrasound-guided intranodal lipiodol LAG from the groin were successfully performed with no complications. Leak points were clearly identified in the three cases. Cure was obtained in one case by the LAG only. Surgical ligations were performed after LAG in two cases and cures were achieved.

DISCUSSION: If LAG successfully cured chylothorax, chest drain output would decrease dramatically and the leaked lipiodol could be confirmed near the leak point in plain computerized tomography (CT) in the following 1–2 days. But if LAG failed to cure, chest drain output would be unchanged and the leaked lipiodol would be found diffusing in the surrounding.

CONCLUSION: Ultrasound-guided intranodal lipiodol LAG from the groin is a minimally invasive and easily performed procedure with high diagnostic and therapeutic value for postoperative chylothorax. If LAG failed to cure, conservative management is often insufficient and surgical ligation should be performed as soon as possible.

© 2016 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Introduction

Postoperative chylothorax are rare complications after esophagectomy, with incidences of approximately 2.7% to 3.8% [1]. Because these conditions can sometimes life threatening if not properly treated, prompt diagnosis and early intervention are necessary. Lipiodol lymphangiography (LAG) has been used in the past to identify sites of chylous. Recent reports also indicate that lipiodol LAG is therapeutic for conventionally untreatable chylothorax [2,3]. Traditionally, lipiodol LAG is performed from pedal lymphatic vessels which are very thin to be identified [4].

Both isolation and cannulation are technically difficult and time-consuming. It also takes time to wait infused lipiodol flowing from pedal lymphatic vessels to groin. Recently, two groups demonstrated the feasibility of using ultrasound-guided intranodal LAG from the groin as an alternative to pedal LAG, achieving success rates of 100% and 87%, respectively [5,6]. The benefit of this novel approach includes reductions in both technical difficulty and procedure duration. But although the proposed benefits of intranodal LAG from the groin may eventually increase the utilization of this novel technique, reports of its use are still few. Herein, we report three cases of post-esophagectomy chylothorax in whom ultrasound-guided intranodal lipiodol LAG from the groin were successfully performed with no complications.

Abbreviations: LAG, lipiodol lymphangiography; CT, computerized tomography; POD, postoperative day; UICC, the Union for International Cancer Control; CDDP, cisplatin; 5-FU, 5-fluorouracil.

⁎ Corresponding author.
E-mail address: yusuke@doc.med.akita-u.ac.jp (Y. Sato).

http://dx.doi.org/10.1016/j.jscr.2016.10.069
2210-2612/© 2016 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).
Fig. 1. (a) Clinical course of case 1. (b) Clinical course of case 2. (c) Clinical course of case 3.
Fig. 2. (a) Ultrasound-guided intranodal LAG demonstrating lipiodol leakage behind the common hepatic arterial trunk (white arrow). (b) Plain CT image obtained after the intranodal lipiodol LAG providing more detailed information about the leak point (white arrow). (c) Plain CT image obtained 1 day after the intranodal lipiodol LAG showing that the leaked lipiodol near the leak point diffused to the surrounding (white arrow). (d) Ultrasound-guided intranodal LAG demonstrating lipiodol leakage at the level of the sixth thoracic vertebra (white arrow). (e) Plain CT image obtained after the intranodal lipiodol LAG providing more detailed information about the leak point (white arrow). (f) Ultrasound-guided intranodal LAG demonstrating lipiodol leakage on the left side of the fifth thoracic vertebra level slightly higher than tracheal bifurcation (white arrow). (g) Plain CT image obtained after the intranodal lipiodol LAG providing more detailed information about the leak point (white arrow). (h) Plain CT image obtained 2 days after the intranodal lipiodol LAG showing remained lipiodol near the leak point (white arrow).
2. Presentation of cases

2.1. Case 1

A 67-year-old man was referred to our hospital for treatment of the Union for International Cancer Control (UICC) cT3N2M0 stage IIIIB esophageal squamous cell carcinoma. Following neoadjuvant chemoradiotherapy, the patient underwent radical esophagectomy with three-field lymph node dissection and resection with a posterior mediastinal gastric tube. Beginning on the 4th postoperative day (POD), the left chest drain output increased to approximating 500–2000 mL/day (Fig. 1a). Because the left chest drain output decreased gradually to 260 mL/day, he was discharged on the 37th POD. However, when the patient visited to our clinic on the 57th POD, he presented with massive accumulation of ascites and left pleural effusion. Upon diagnostic left thoraecentesis, milky pleural fluid was collected, confirming the diagnosis of left chylothorax. Initially, we treated the patient conservatively, providing total parenteral nutrition and administering octreotide for 2 weeks (0.5 mg, 3 times a day, subcutaneously). However, the left chest drain output continued to range between 500 and 1500 mL/day. On the 93rd POD, we performed ultrasound-guided intranodal lipiodol LAG from the right groin (LAG time: 1 h 6 m 48s, lipiodol 8 mL) and successfully detected leakage of lipiodol from behind the common hepatic arterial trunk (Fig. 2a, b). Although previous reports indicate that chylothorax can be cured with lipiodol LAG [3], unfortunately, in this case, there was no reduction in pleural effusion. Plain CT image obtained 1 day after the intranodal lipiodol LAG showed that the leaked lipiodol near the leak point diffused to the surrounding (Fig. 2c). On the 106th POD, we performed a surgical ligation of the leaking lymph vessel. After the surgery, the output decreased, and we removed both of the left chest drain and abdominal drain on the 122nd POD and discharged the patient on the 136th POD. Subsequent examinations in our clinic showed no accumulation of ascites or pleural effusion in this patient.

2.2. Case 2

A 59-year-old man was referred to our hospital for treatment of UICC cT3N1M0 stage IIIA esophageal squamous cell carcinoma. Following neoadjuvant chemoradiotherapy, the patient underwent robot-assisted minimally invasive esophagectomy with three-field lymphadenectomy and reconstruction with a posterior mediastinal gastric tube. Beginning on the 2nd POD, the left chest drain output increased to approximately 1000–2500 mL/day (Fig. 1b). Milky pleural fluid was observed soon after the milk intake on the 10th POD, confirming the diagnosis of left chylothorax. Because there was massive fluid accumulation beneath the cervical wound, a thoracic duct injury in the cervical region was suspected. We therefore reopened the left cervical wound and applied clips to suspicious potential leak points in the thoracic duct. Nonetheless, the left chest drain output remained over 1200 mL/day. On the 12th POD, we performed ultrasound-guided intranodal lipiodol LAG from the right groin (LAG time: 1 h 3 m 18s, lipiodol 15 mL), which was followed with plain CT. Lipiodol leakage was detected on the left side at the level of the sixth thoracic vertebra (Fig. 2d, e). The following day, the left chest drain output was still 1375 mL, indicating that lipiodol LAG did not stop the chylosus leakage in this patient either. Given the high output and successful detection of the leak point, on the 14th POD we performed surgical ligation of the thoracic duct. Thereafter, the left chest drain output decreased significantly, and the drain was removed 2 days later. The patient was discharged on the 22nd POD.

2.3. Case 3

A 75-year-old man was referred to our hospital for treatment of UICC cT1N1M0 stage IIIB esophageal squamous cell carcinoma. The patient underwent thoracoscopic esophagectomy with three-field lymphadenectomy and reconstruction with a posterior mediastinal gastric tube. Beginning on the 4nd POD, the right chest drain output increased to approximately 1000–2000 mL/day (Fig. 1c). Initially, we treated the patient conservatively, providing total parenteral nutrition and administering etilefine hydrochloride for 9 days (100–120 mg/day, continuous infusion) [7]. However, the right pleural effusion did not reduce. On the 21st POD, we performed ultrasound-guided intranodal lipiodol LAG from the right groin (LAG time: 40m, lipiodol 8 mL) which was followed with plain CT. We successfully detected leakage of lipiodol on the left side of the fifth thoracic vertebral level slightly higher than tracheal bifurcation (Fig. 2f, g). The following day, the right chest drain output dramatically decreased to 292 mL. On the 23rd POD, plain CT showed that leak point was likely still being blocked by the remained leaked lipiodol (Fig. 2h). Thereafter, the right chest drain output did not increase and the drain was removed on the 41st POD. Because of postoperative swallowing disturbance, the patient suffered repeated aspiration pneumonitis and was discharged on the 70th POD.

3. Discussion

Traditionally, lipiodol LAG entails a bilateral pedal approach that includes isolation and cannulation of pedal lymphatic vessels followed by infusion of lipiodol [5]. Since Nadolski and Itkin demonstrated the feasibility of intranodal LAG as an alternative to pedal LAG in 2013 [6], ultrasound-guided intranodal lipiodol LAG from the groin has been tried in some facilities [7]. Because inguinal lymph nodes are easy to be identified under ultrasound guidance, and because isolation of lymphatic vessel will not be necessary, ultrasound-guided intranodal lipiodol LAG from the groin has been shown to be a minimally invasive and easily performed procedure. Reports published up to now suggest that it is a reliable, reproducible technique without major complications [8]. In our 3 cases, patients were placed in a prone position on fluoroscopy units (Artis Zee, Siemens Medical Solutions, Erlangen, Germany). Local anesthesia of the puncture site was induced with 1% lidocaine, after which the inguinal lymph node was punctured under ultrasound guidance using a 23-gauge needle, the tip of which was placed at the junction between the cortex and the hilum. Lipiodol (Terumo, Tokyo, Japan) injection was performed manually at a rate of 0.1 mL/min. During the injection, fluoroscopy was used intermittently to confirm that the injected lipiodol was entering the lymphatic vessels [9].

Lipiodol LAG has been proven useful for diagnosis of postoperative lymphatic leakage, and recent reports suggest lipiodol LAG is also therapeutic for conventionally untreatable chylothorax [2,3]. It was hypothesized that extravascular lipiodol induces a granulomatous reaction that can result in closure of the leak [3]. In fact, therapeutic benefit was obtained in our case 3. Whether therapeutic benefit is obtained or not could be related to the size of leak, the chylous output or the amount and density of lipiodol used. Further study is still needed.

The timing of LAG for postoperative chylothorax is an important issue that should be considered. Undiagnosed postoperative chylothorax may lead to malnutrition, sepsis and even death, making prompt diagnosis and early intervention are important. A continuous output in excess of 1000 mL/day may be a good indicator of LAG [8], but the definitive duration of high output is still being debated.
From our 3 cases, we found that if lipiodol successfully cured the chylothorax, chest drain output would decrease dramatically and the leaked lipiodol could be confirmed near the leak point in plain CT in the following 1–2 days. But if lipiodol failed to cure, chest drain output would be unchanged and the leaked lipiodol would be found diffusing into the surrounding. In the latter situation, conservative management is often insufficient and surgical ligation should be performed as soon as possible.

4. Conclusion

Ultrasound-guided intranodal lipiodol LAG from the groin is a minimally invasive and easily performed procedure with high diagnostic and therapeutic value for postoperative chylothorax. We recommend an early performance of this procedure. Then depending upon whether chest drain output decrease and whether leaked lipiodol is still remaining near the leak point in plain CT performed 1–2 days after LAG, the next therapeutic strategy can be decided more reasonably.

Conflicts of interest

All authors state that they have no financial competing interests to disclose.

Authors’ contributions

J.L. and Y.S. made substantial contributions to acquisition of data and to draft the manuscript. S.M., K.L., K.Y., M.Y. participated in its design and coordination and helped to draft the manuscript. A.W., Y.K., K.I., H.S., M.H. and Y.M. participated in the design of the study.

Ethical approval

This study was approved by the Ethics Committee of Akita University School of Medicine. All of the participants provided informed consent and signed a human subject institutional review board consent form.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

SCARE Guideline

The work is compliant with the SCARE Guideline [10].

Guarantor

Yusuke Sato and Satoru Motoyama.

Funding

This work was not supported by any foundation.

References

[1] R.D. Shah, J.D. Luketich, M.J. Schuchert, et al., Postesophagectomy chylothorax: incidence, risk factors, and outcomes, Ann. Thorac. Surg. 93 (2012) 897–903, discussion 903-894.
[2] M. Yamamoto, H. Miyata, M. Yamasaki, et al., Chylothorax after esophagectomy cured by intranodal lymphangiography: a case report, Anticancer Res. 35 (2015) 891–895.
[3] S. Kos, H. Hauesen, U. Lachmund, et al., Lymphangiography: forgotten tool or rising star in the diagnosis and therapy of postoperative lymphatic vessel leakage, Cardiovasc. Intervent. Radiol. 30 (2007) 968–973.
[4] C. Cope, L.R. Kaiser, Management of unremitting chylothorax by percutaneous embolization and blockage of retroperitoneal lymphatic vessels in 42 patients, J. Vasc. Interv. Radiol. 13 (2002) 1139–1148.
[5] G.J. Nadolski, M. Itkin, Feasibility of ultrasound-guided intranodal lymphangiogram for thoracic duct embolization, J. Vasc. Interv. Radiol. 23 (2012) 613–616.
[6] A. Parvinian, G.C. Mohan, R.C. Gaba, et al., Ultrasound-guided intranodal lymphangiography followed by thoracic duct embolization for treatment of postoperative bilateral chylothorax, Head Neck 36 (2014) E21–24.
[7] F. Guillem, V. Billerot, M.L. Houcke, et al., Successful management of post-esophagectomy chylothorax/chyloperitoneum by etilefrine, Diseases Esophagus 12 (1999) 155–156.
[8] R. Bender, V. Murthy, R.S. Chamberlain, The changing management of chylothorax in the modern era, Eur. J. Cardiothorac. Surg. (2015).
[9] S. Kariya, A. Komemushi, M. Nakatani, et al., Intranodal lymphangiogram: technical aspects and findings, Cardiovasc. Intervent. Radiol. 37 (2014) 1606–1610.
[10] R.A. Agha, A.J. Fowler, A. Saeta, et al., The SCARE Statement: consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180–186.