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Case report

Intranodal lymphangiography under microsurgery for refractory lymphatic ascites after pelvic lymphadenectomy

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ABSTRACT

Lymphatic ascites is a postoperative complication of lymph node dissection. Most symptomatic cases improve with conservative treatments. However, optimal management strategies for intractable lymphatic ascites remain controversial, and clinicians sometimes encounter intractable lymphatic ascites that does not respond to conservative management.

We herein report a case of postoperative intractable lymphatic ascites that was successfully treated with intranodal lymphangiography (LG) from inguinal lymph nodes under microsurgery. A 56-year-old woman was diagnosed with stage II endometrial cancer and underwent total abdominal hysterectomy, bilateral salpingo-oophorectomy, and pelvic and *para*-aortic lymphadenectomies. On postoperative day (POD) 13, the patient presented with abdominal distention, and lymphatic ascites was diagnosed. Although the patient was treated with conservative management and lymphaticovenular anastomosis, her lymphatic ascites did not resolve. Finally, intranodal LG from the inguinal region was performed under microsurgery. A 2-cm incision was made on each side of the inguinal region. Once the lymph nodes were identified, a 23-gauge needle was inserted into the lymph node and lipiodol was injected. Extravasation of lipiodol into the abdomen from the left side of the lower pelvic region was confirmed. The postoperative course was uneventful. The ascites gradually decreased and disappeared within two weeks after LG.

1. Introduction

Lymphatic ascites is a postoperative complication of lymph node dissection, and its incidence in gynecologic oncology is reported to be 4 % (Frey et al., 2012). Most symptomatic cases improve with conservative treatment, including dietary modification and paracentesis. However, clinicians sometimes experience intractable lymphatic ascites that does not respond to conservative management (Frey et al., 2012; Krishnan et al., 2001). This may delay adjuvant treatment and increase the risk of recurrence (Zhu et al., 2021). Therefore, immediate improvement of the refractory ascites is required.

The optimal management strategies for intractable lymphatic ascites remain controversial. Lymphangiography (LG) is a potentially effective treatment for lymphatic ascites (Lee et al., 2014; Baek et al., 2016). LG is a technique for injecting contrast agents, such as lipiodol, from lymphatic ducts and lymph nodes to identify and treat lymphatic leakage. The therapeutic effect is attributed to the embolic properties of lipiodol (Lee et al., 2014). There are two types of LG: pedal and intranodal. The pedal LG first needs to identify the lymphatic duct by injecting indigo carmine from the toe. Once the lymphatic duct is found, it is isolated the lymphatic duct with a small incision, and a contrast agent is injected using a 30-gauge LG needle. These techniques require training and devices (Lee et al., 2014). Intranodal LG is also a minimally invasive procedure, and the largest and most distal inguinal lymph nodes are directly accessed under ultrasound guidance (Lee et al., 2014). More than 80 % of lymphatic leakage cases were successfully treated with ultrasound-guided intranodal LG (Baek et al., 2016). However, physicians sometimes encounter technical difficulties in pedal LG and

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ultrasound-guided intranodal LG, especially when the lymph nodes are too small to puncture.

We herein report a case of postoperative intractable lymphatic ascites that was successfully treated with intranodal LG from the inguinal lymph nodes under microsurgery.

2. Case report

A 56-year-old woman was diagnosed with stage II endometrial cancer and underwent total abdominal hysterectomy, bilateral salpingo-oophorectomy, and pelvic and *para*-aortic lymphadenectomies. The intraoperative blood loss was 425 ml. The number of the resected lymph nodes of left pelvic, right pelvic and *para*-aortic were 22, 16, and 17, respectively. The patient was discharged from the hospital uneventfully. On postoperative day (POD) 13, the patient presented with abdominal distention. Abdominal computed tomography (CT) showed massive ascites of unknown origin (Fig. 1a). Abdominal paracentesis was performed, and the ascites was pale yellow. A laboratory examination of the ascites revealed no evidence of chylous ascites, bacterial infection, and urine leakage into the abdominal cavity. The triglyceride and cholesterol levels were normal, but the differential count of leukocytes was lymphocyte-predominant. Lymphatic ascites was diagnosed.

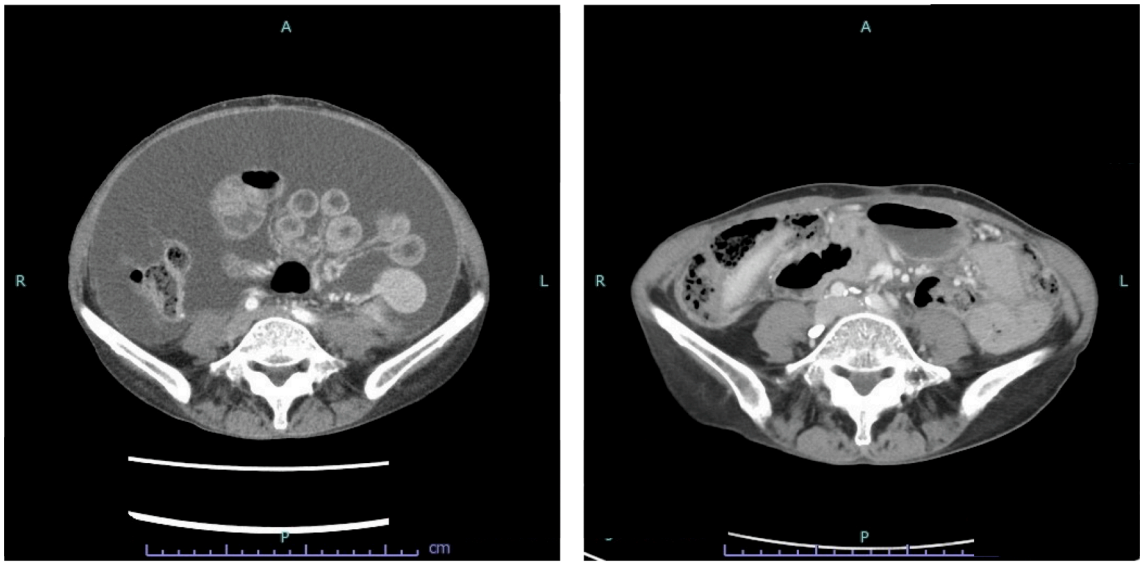
The clinical course of the patient is shown in Fig. 2. Although the patient was treated conservatively, including total parenteral nutrition without oral intake and octreotide therapy for the first two weeks, the lymphatic ascites did not resolve. Interventional treatments were then proposed, but periodic paracentesis was performed every one or two weeks at the patient's request. Periodic paracentesis was performed for 3 months, and the ascites did not decrease. The patient agreed to undergo interventional therapy.

Three interventional treatment options were considered: ultrasound-guided LG, lymphaticovenular anastomosis (LVA), and ligation of lymphatic leakage by surgery. Although minimally invasive therapy was preferred, ultrasound-guided intranodal LG from the inguinal lymph nodes or pedal LG was difficult to perform because the lymph node was too small to puncture. Therefore, LVA was performed on POD 110 and 159 in this case. Before LVA, indocyanine green (ICG) lymphography was performed to find which lymphatic duct and vein to be anastomosed. Total of eleven vessels and lymphatic ducts on both sides of the lower limbs were anastomosed; however, the amount of ascites did not decrease. Finally, we decided to perform intranodal LG in the inguinal region via microsurgery on POD 205.

Intranodal LG from the inguinal region was performed under general anesthesia. A 2 cm incision was made parallel to the inguinal ligament and above the femoral vessels on both sides. (Fig. 3a). Loose connective and adipose tissues around the lymph nodes were carefully removed to avoid injuring the lymphatic duct. Once the lymph node (approximately 5 mm in size) was identified, a 23-gauge needle was inserted into the lymph node, and lipiodol was injected (Fig. 3b and c). Extravasation of lipiodol into the abdomen from the left side of the lower pelvic region was confirmed (Fig. 3d and Supplementary material). We also injected lipiodol to the lymph node of the right inguinal region. There was no sign of lymphatic leakage in the right pelvic or *para*-aortic region. The postoperative course was uneventful. The ascites gradually decreased and disappeared within two weeks after LG. On postoperative day 46 after LG, there was no evidence of ascites on the CT scan (Fig. 1b). She was completely symptom-free after LG, and there was no disease recurrence during 14 months of postoperative follow-up.

3. Discussion

A small amount of lymphatic ascites may occur after lymphadenectomy for gynecological surgery; however, ascites usually resolves spontaneously without the development of symptoms. Lymphatic ascites is straw-colored or clear fluid with low contains of triglyceride, which differs from chylous ascites (milky fluid with high triglyceride levels). The reported incidence of postoperative symptomatic lymphatic ascites in patients undergoing gynecologic malignancies is 2.7–4.0 % (Frey et al., 2012; Krishnan et al., 2001). However, most cases were improved by dietary modifications and paracentesis and did not require further treatment. The interval between the diagnosis and resolution was less than 1 month in most cases and no longer than 3 months in others (Frey et al., 2012; Krishnan et al., 2001). If lymphatic ascites persists, further cancer treatment may be delayed. Delayed adjuvant treatment increases the risk of recurrence (Zhu et al., 2021), therefore the immediate improvement of ascites is required. Several management options for interactable lymphatic ascites, including LG, LVA, or direct repair of retroperitoneal lymph vessel injuries, should be considered (Lee et al., 2014; Baek et al., 2016; Liu et al., 2018; Todokoro et al., 2013; Hattori et al., 2022). However, the optimal treatment strategies remain



(a) (b)

Fig. 1. Computed tomographic images of patient. (a) Before lymphangiography, the patient had massive ascites. (b) After lymphangiography, the ascites significantly improved.

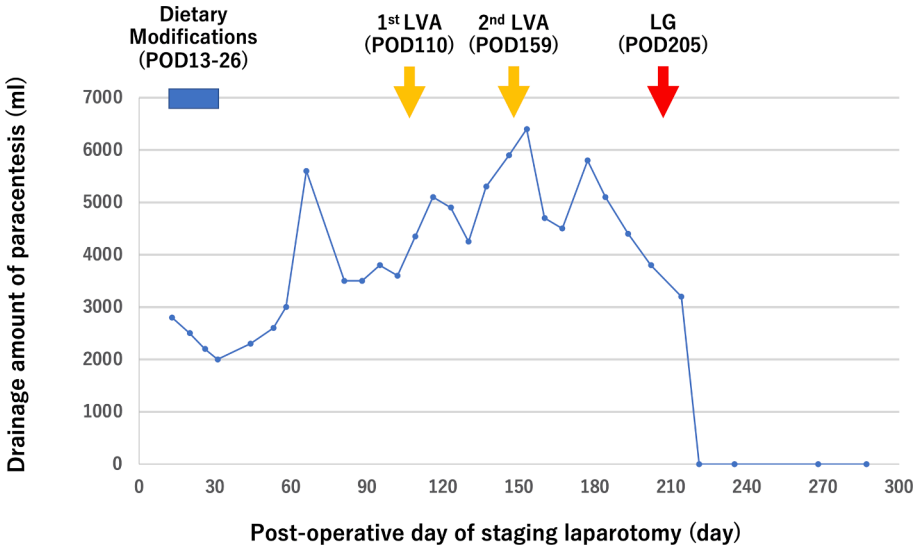


Fig. 2. Amount of paracentesis drainage and timing of interventions LVA: Lymphaticovenular anastomosis, LG: Lymphangiography, POD: Postoperative day.

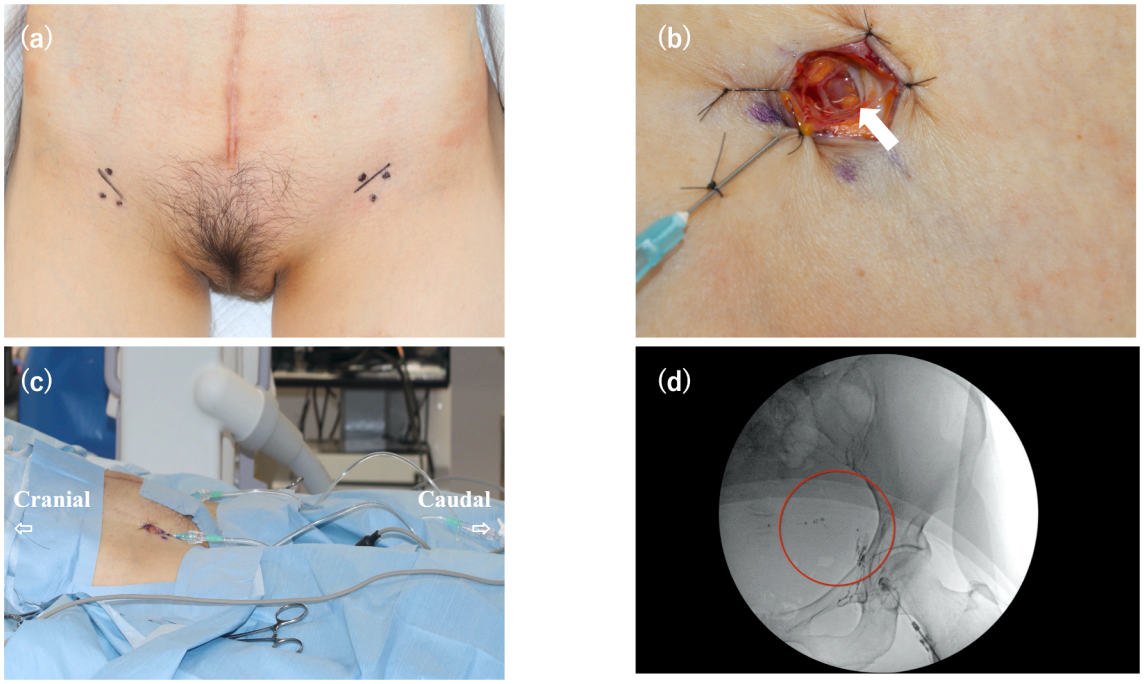


Fig. 3. Intranodal lymphangiography from the inguinal lymph node under microsurgery. (a) Incision lines of the inguinal region. (b) Exposure of the lymph node under microsurgery and the inguinal lymph node puncture with a 23-G needle. The white arrow indicates exposed lymph node. (c) Administration of contrast agent to the inguinal lymph node. (d) Image of lymphangiography. The red circle indicates leakage of contrast agent. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

unknown because there are still unresolved controversies regarding the appropriate selection of candidates, and evidence comparing the efficacy of these procedures is lacking. Furthermore, it depends on resource availability and physician's preferences.

LVA is a minimally invasive treatment that is becoming increasingly popular for the treatment of lymphedema. LVA allows anastomosis with the proximal lymphatic duct and venous system to provide an alternative route for lymphatic fluid recirculation into the venous system. Recently, there have been reports on the use of the LVA technique for the treatment of lymphatic leakage (Todokoro et al., 2013; Hattori et al., 2022). Several anastomoses of the lymphatic duct and vein in both lower limbs decrease the lymphatic flow into the abdominal cavity. Although we performed LVA prior to LG, we failed to reduce lymphatic flow. Unfortunately, LVA has not proven to be effective in all patients with lymphatic ascites (Mihara et al., 2015). Furthermore, it is difficult to predict the extent to which one LVA reduces lymphatic flow at the injured site preoperatively, and there is uncertainty regarding the exact number of LVAs that should be placed during a procedure to ensure adequate treatment (Kadota et al., 2021).

LG is an effective method for detecting lymphatic leakage by injecting contrast agents, such as lipiodol, from lymph nodes or lymphatic ducts. Lipiodol is ethiodized oil, which retains in the lymphatic system and induces sterile inflammatory reaction to block the lymphatic ducts. According to previous studies, LG plays a therapeutic role and is successful in 75 % of lymphatic leakage cases (Lee et al., 2014). Recently, ultrasound-guided intranodal LG from the inguinal

region has become popular owing to its minimal invasiveness. Although ultrasound-guided intranodal LG has a relatively high success rate (Baek et al., 2016), there are technical difficulties when the inguinal lymph nodes are too small to puncture. Furthermore, pedal LG is contraindicated when LVA is performed prior to LG because contrast agents such as lipiodol cause pulmonary or cerebral embolism when the drug flows into the vein (Lee et al., 2014). Therefore, in our case, injection of the contrast agent from the proximal side of the anastomosis of the lymphatic duct and vein was necessary. Based on the size of the inguinal lymph nodes and previous history of LVA in the lower limbs, intranodal LG from the inguinal region under microsurgery was considered a reasonable approach.

To inject the contrast agent directly, the lymph nodes should be exposed without injuring the lymphatic duct. Because lymphatic ducts are very thin (with diameters of 0.4–0.6 mm) and are surrounded by fatty tissues, they are easily damaged during lymph node exposure (Hara and Mihara, 2021). Therefore, microsurgical techniques enable the identification of lymph nodes with less injury to the lymphatic ducts. Although there is no prospective study of LG under microsurgery and the evidence regarding intranodal LG under microsurgery is limited, it might be an alternative method, especially when the size of lymph nodes is too small, when ultrasound-guided intranodal LG fails, or when lymphatic ascites does not respond to LVA.

In conclusion, intranodal LG under microsurgery is an effective method for treating refractory lymphatic ascites, and could be a treatment option for this complication.

4. Consent to participate

Informed consent was obtained from the patient.

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None.

CRediT authorship contribution statement

Shota Higami: Writing – original draft, Investigation, Conceptualization. **Yusuke Tanaka:** Writing – review & editing, Writing – original draft, Conceptualization. **Daisuke Maeda:** Writing – review & editing. **Hiroshi Yukimoto:** Writing – review & editing. **Tomomi Ishii:** Writing – review & editing. **Yasuhiko Shiki:** Writing – review & editing, Supervision.

Data availability

All of the material used during the manuscript are available from the corresponding author on reasonable request.

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Not Applicable.

Appendix A. Supplementary material

Supplementary data to this article can be found online at https://doi.org/10.1016/j.gore.2024.101346.

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