IMR Press

European Journal of Gynaecological Oncology

Review

Interventional therapy of refractory lymphatic ascites post pelvic lymphadenectomy: a case report and literature review

Xiao-Yu Zhang[1], Yang Shen[1,*]

[1] *Department of Obstetrics and Gynecology, Zhongda Hospital, Southeast University, 210009 Nanjing, China*

*Correspondence: shenyang0924@sina.cn (Yang Shen)

DOI:10.31083/j.ejgo.2021.02.2202

Submitted: 25 July 2020 Revised: 11 September 2020 Accepted: 14 September 2020 Published: 15 April 2021

Lymphatic ascites is an infrequent but severe complication that can occur after lymphadenectomy in a patient who had gynecologic malignancies. Without appropriate management, lymphatic ascites can induce electrolyte disorder, hypoproteinemia, immunologic dysfunction, and even life-threatening. However, the optimal management is still unknown. We report a case with successful management of refractory pelvic lymphatic ascites in a patient with cervical cancer status post laparoscopic radical hysterectomy and pelvic lymph node dissection (PLND). Ultrasound-guided intranodal lymphangiography (UGILG), an innovative procedure, was used, and a leakage point at the left iliac fossa was identified after unsuccessful conservative treatments. Leakage of lymphatic fluid was treated successfully by embolizing the defect area under the guide of UGILG. UGILG is a feasible and alternative procedure for the diagnosis and treatment of lymphatic ascites, especially in women with a complicated gynecological procedure.

Keywords

Lymphangiography; Lymphadenectomy; Lymphatic leakage; Treatment

1. Introduction

Lymphatic ascites is one of the postoperative complications in gynecologic malignancies, which we need to pay more attention to. Lymphatic ascites can induce electrolyte disorder, hypoproteinemia, immunologic dysfunction, and even life-threatening without appropriate management [1–3]. However, optimal management is still unknown [4]. We report a case with successful management of refractory pelvic lymphatic ascites in a patient with cervical cancer status post laparoscopic radical hysterectomy and pelvic lymph node dissection by using UGILG.

2. Case presentation

A 43-year-old female was diagnosed to have FIGO stage IB1 cervical cancer when she had her annual gynecology checkup. Her past medical history is ordinary and unremarkable. She had a standard laparoscopic radical hysterectomy and pelvic lymph node dissection (PLND) as per the recommendation of FIGO 2018 on 06 November 2018.

The surgery was performed successfully without any noted complications in 182 minutes with about 100mL blood losses, and a pelvic drainage tube was placed as routine. On the post-operative day (POD) 1, greater than 1000–1500 mL of clear yellowish pelvic drainage was noted. Further evaluations were performed, and the urinary system injuries or complications were ruled out including serum and drainage creatinine level, the intravesical methylene blue perfusion test of the bladder were all within the normal range (lab analysis of the ascitic fluid: gross appearance: faint yellow; specific gravity: 1.012; total protein: 30 g/L; triglyceride: 111 mg/dL; red blood cell: 200 $\times$ 106/L; white blood cell: 1000 $\times$ 106/L; bacteria: Not detected; creatinine: 20 μmol/L). Therefore, post-operative refractory lymphatic ascites (PRLA) were considered. Conservative management as routine, four including medium-chain triglyceride diet and total parenteral nutrition (TPN), as well as medical administration such as somatostatin analogs, diuretics, was initiated the same day of post-operative day one. Even after the initiation of conservative management immediately after the clinical diagnosis of PRLA, it appeared to have minimal effects. Pelvic drainage continued having 1100–1300 mL clear yellowish fluid daily. In POD#6, the patient complained of suddenly a large volume of vaginal clear yellowish discharge. Vaginal examination showed left vaginal cuff dehiscence with clear yellowish discharge that was the same as pelvic drainage. Conservative management was continued until POD#14. The patient still complained of continuous vaginal discharge, and her general condition and nutritional status were getting worse with hypoproteinemia. We considered the failure of conservative methods because of the constant drainage of large amounts of fluid. Therefore, interventional radiology was consulted, and an ultrasound-guided intravascular lymphangiography (UGILG) was recommended that was described as routine procedures in detail as following.

Step 1. Ultrasonic examination detected enlarged multiple enlarged lymph nodes in the left inguinal region. One deep lymph node at left groins was selected out as the target lymph node, which was punctured with a 25-gauge ultra-fine needle under the guidance of ultrasound (Fig. 1A–C).

Step 2. Lymphography was performed using an intranodal technique using digital subtraction angiography (DSA) system with contrast injection to lymph nodes at both inguinal

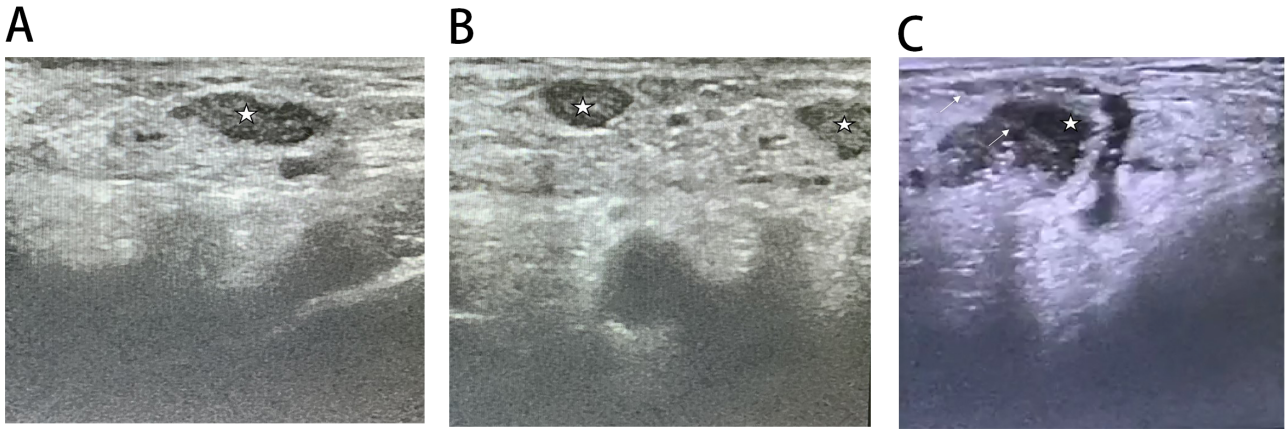

Fig. 1. (A,B) "⋆" shows the left inguinal lymph node. (C) "⋆" shows the puncture approach for target lymph node, and "→" shows the puncture needle entering the lymph node accurately.

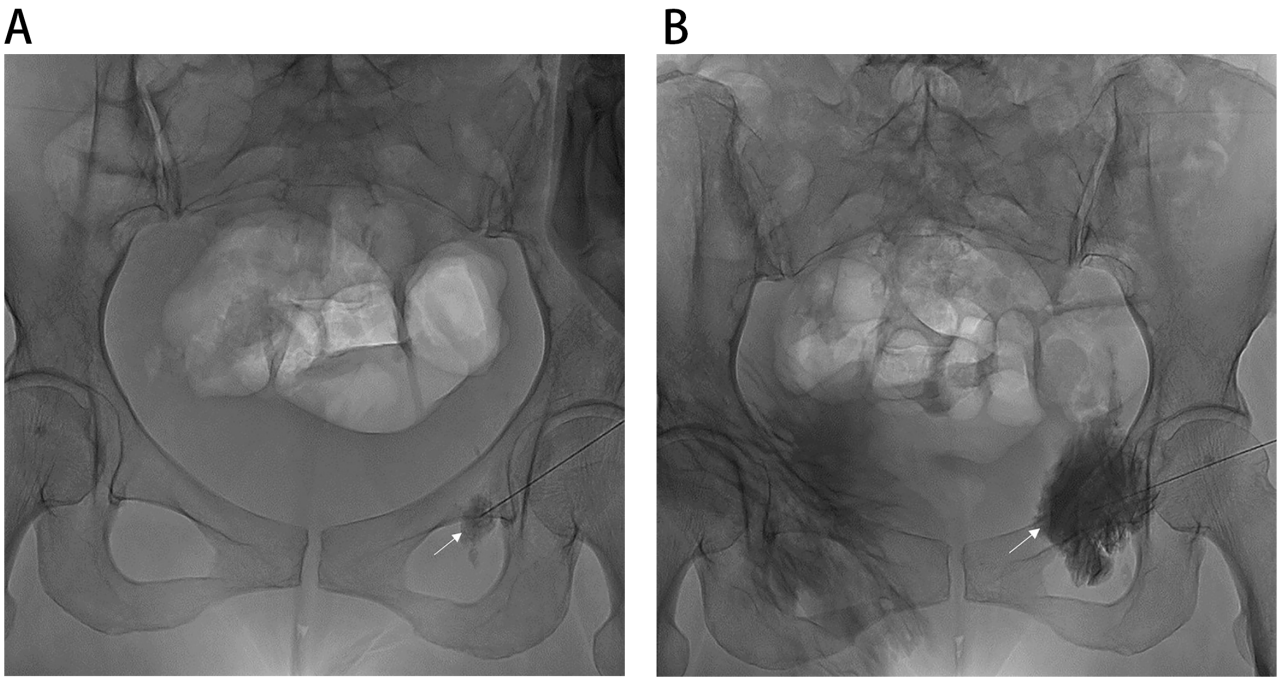

Fig. 2. (A) The lipiodol was slowly injected into the target lymph node to display the mapping of lymphatic vessels. (B) It shows a lymphatic vessel at the left iliac fossa enlarged like multi fusiform lymphatic aneurysms (→).

regions (Fig. 2A). The 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. We found lymphatic lesion at the left iliac fossa when lymphatic vessels were opacified. Finally, a lymphatic vessel at the left iliac fossa was found to be enlarged like multi fusiform lymphatic aneurysms (Fig. 2B).

Step 3. The imaging indicated that the lymphatic fluid leakage into the abdomen cavity with the site of lymphatic leakage was recognized. Embolization of defect was performed with injecting 1 mL mixture of n-butyl cyanoacrylate (NBCA), which was diluted with lipiodol at the ratio 1 : 2, through the puncture needle into the lymphatic vessels (Fig. 3).

After the procedure, the amount of pelvic drainage and vaginal leaking decreased significantly on the same day as the procedure and continue decreasing significantly every day. On post embolism day 3, only minimal pelvic drainage and vaginal leakage left (Fig. 4). Pelvic drainage was wholly removed on post embolism day 5.

The pathology report showed stage IB1 cervical squamous carcinoma, and no adjuvant post-operative chemotherapy or radiation was indicated.

The patient was discharged home on POD#20 with a weekly phone visit, and the patient was scheduled to have a clinical visit in two weeks. Vaginal stump healed well in 2 months without any other intervention.

Fig. 3. It showed a large amount of iodide oil leaking into the pelvic cavity, which is the lymphatic leakage site. Embolization was performed by injecting NBCA through the puncture needle into the lymphatic vessel.

3. Discussion

As we all know, pelvic and para-aortic lymph node dissection (PLND) is a significant component of the surgical staging procedure for several gynecologic malignancies, including endometrial, cervical, and ovarian carcinoma [5]. The results of PLND are pivotal to evaluate the prognosis as well as guide the adjuvant therapy of patients. However, there are some rare but potentially life-threatening complications associated with PLND, including traumatic lymphatic ascites. During operation, the trauma of the lymphatic system leads to the post-operative leakage of massive lymphatic fluid. In most cases, it usually ceases spontaneously without the consequence of symptomatic ascites, and only 4% of the gynecological malignancies are identified with symptoms and complaints, which are most commonly reported as leakage of clear fluid per vagina (58%) and abdominal distension (33%). The median interval from surgery to the development of symptoms ranges from 0 to 22 days [6]. The present case was confirmed by the vast volume of pelvic drainage of lymphatic fluid on POD#1 and consequent vaginal leakage of lymphatic fluid on POD#6.

What about the possible causes of lymphatic ascites? First, in laparoscopic surgery, it was inevitable to use the electrocoagulation function of energy devices, such as bipolar coagulation and ultrasonic scalpel. During the operation, these energy devices may only temporarily close the rupture of lymphatic vessels. However, when pneumoperitoneum was removed, the abdominal pressure reduced, while the pressure in lymphatic vessels increased, lymphatic fluid could break through the eschar of the electrocoagulation wound and leak into the abdominal cavity, resulting in lymphatic ascites. Second, the patient started to take a semi-liquid diet on POD#2. With the fat intake, the volume and pressure in the lymphatic vessels increased, which may contribute to the occurrence of lymphatic ascites.

Once lymphatic ascites was diagnosed, treatment should be started immediately to avoid the possible life-threatening complication such as hypoproteinemia and severe infection. Until now, there is still no consensus or standard guideline for the management of lymphatic ascites, especially for refractory cases. 70% of patients can be successfully treated by conservative methods, which consists of percutaneous catheter drainage and parenteral nutrition to decrease the production of lymphatic fluid until the damaged lymphatic channel spontaneously heals [7]. Meanwhile, diet control, nutrition support, and application of some drugs, such as somatostatin analogs, diuretics *et al.*, are all feasible approaches [8, 9]. After 4–6 weeks of conservative treatment, if the drainage volume was still greater than 500 mL per day, it is generally believed that the patient suffered refractory lymphatic ascites. Then, aggressive management, including interventional and surgical treatment, should be considered for refractory cases. Compared to surgical management, the interventional treatment appeared to be an optimal alternative for lymphatic ascites [10–12]. The surgical procedure, in general, is recommended as a final approach, including peritoneovenous shunt (PVS), direct lymphostasis by suture ligation of the disrupted lymphatic vessels, or surgery combining with fibrin glue [13]. It is frequently challenging to detect the site of lymphatic leakage intraoperatively. A novel technique of surgical intervention is laparoscopic management using fluorescence navigation with indocyanine green. Intraoperative indocyanine green injection is useful for detecting a lymphatic leakage site and especially making sure without other leakages [14].

From our experience, the innovative technique of "closet upstream lymph node embolization" for interventional therapy should be considered before surgery to avoid potential surgical complications. The closest upstream lymph node was identified as the lymph node from which efferent lymphatic vessels were extravasated. Lymphography can be used to map through inguinal lymph nodes to identify the area of leaking. When a leaking area was found, lymphatic embolization should be considered if applicable by fine-needle injection of the mixture of N-butyl cyanoacrylate, which was diluted with lipiodol at the various ratio (range 1 : 2–1 : 8), into a pelvic or inguinal lymph node located below the leakage [15, 16]. In this case, we choose unilateral other than bilateral inguinal lymph nodes puncture because that lymphatic leakage site usually determines in the single lateral of the body, most possibly in the lateral of enlarged groin lymph nodes resembling the lymphatic obstruction. Retrospective studies reported the effectiveness and safety of lymphography and embolization in the treatment of lymphatic ascites after PLND. The overall success rate was 85–100%. The mean

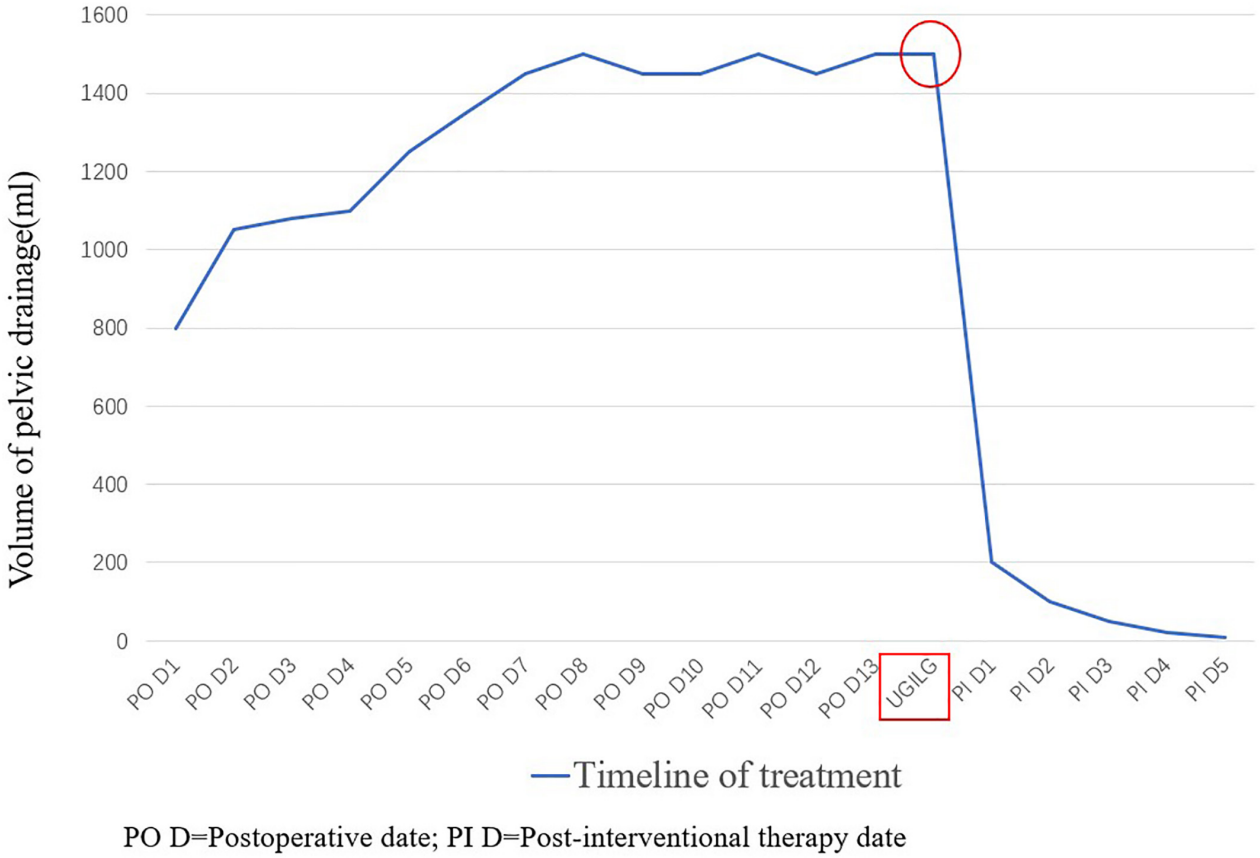

Fig. 4. The volume of pelvic drainage plus vaginal discharge rapidly decreased at the first day after UGILG.

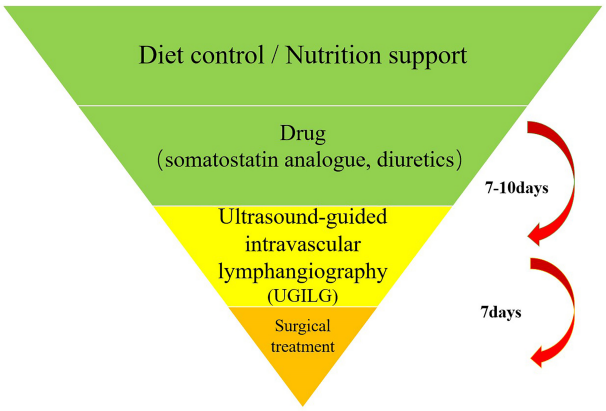

Fig. 5. The treatment algorithm of lymphatic ascites.

time for recovery after the lymphatic intervention was 5–7 days [17, 18]. The complications of UGILG may include prolonged hospitalization, infection, hemorrhage, mild transient leg edema, which could be managed conservatively [19].

Based upon our experience, we propose an algorithm of management of lymphatic ascites as following (Fig. 5). Conservative therapy is the first step of management, and UGILG is recommended for refractory patients after 7–10 days of conventional therapy. The clinical success rate of lymphangiography and adjunctive embolization is 85%, with no significant complications reported. The second session lymphatic vessel embolization could be performed one week after the first session because of persistent leakage [16]. Surgical treatment is considered as the last step after the failure of interventional treatment.

Lymphatic ascites is a rare but possible life-threatening post-operative complication of PLND. Most patients are asymptomatic and could be managed conservatively. UGILG should be considered as an applicable alternative procedure for the failure of conservative therapy for refractory cases before surgery.

Author contributions

XYZ wrote the manuscript. YS revised the manuscript critically for relevant intellectual content and approved the version of the manuscript to be published. Both authors contributed to editorial changes in the manuscript. Both authors read and approved the final manuscript.

Ethics approval and consent to participate

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

Acknowledgment

Not applicable.

Funding

This study was supported by the National Natural Science Foundation of China (No. 81673130).

Conflict of interest

The authors declare no conflict of interest.

References

[1] Pan W, Yang C, Cai S, Chen Z, Cheng N, Li F, *et al.* Incidence and risk factors of chylous ascites after pancreatic resection. International Journal of Clinical and Experimental Medicine. 2015; 8: 4494–4500.

[2] Hara H, Mihara M, Hayashi A, Kanemaru M, Todokoro T, Yamamoto T, *et al.* Therapeutic strategy for lower limb lymphedema and lymphatic fistula after resection of a malignant tumor in the hip joint region: a case report. Microsurgery. 2014; 34: 224–228.

[3] Tiong HY, Goel RK, White WM, Goldfarb DA, Kaouk JH. Chylous ascites after laparoscopic donor nephrectomy. Asian Journal of Endoscopic Surgery. 2015; 8: 34–39.

[4] 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. The Journal of Obstetrics and Gynaecology Research. 2015; 40: 622–626.

[5] Lee YS. Single-port laparoscopic transperitoneal infrarenal paraaortic lymphadenectomy as part of staging operation for early ovarian cancer and high grade endometrial cancer. Journal of Gynecologic Oncology. 2016; 27: e32.

[6] Frey MK, Ward NM, Caputo TA, Taylor J, Worley MJ, Slomovitz BM. Lymphatic ascites following pelvic and paraaortic lymphadenectomy procedures for gynecologic malignancies. Gynecologic Oncology. 2012; 125: 48–53.

[7] Lorenz K, Abuazab M, Sekulla C, Nguyen-Thanh P, Brauckhoff M, Dralle H. Management of lymph fistulas in thyroid surgery. Langenbeck's Archives of Surgery. 2011; 395: 911–917.

[8] Weniger M, D'Haese JG, Angele MK, Kleespies A, Werner J, Hartwig W. Treatment options for chylous ascites after major abdominal surgery: a systematic review. American Journal of Surgery. 2016; 211: 206–213.

[9] Creavin B, Ryan E, Martin ST, Hanly A, O'Connell PR, Sheahan K, *et al.* Organ preservation with local excision or active surveillance following chemoradiotherapy for rectal cancer. British Journal of Cancer. 2017; 116: 169–174.

[10] 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. Journal of Radiology Case Reports. 2020; 15: 1623–1628.

[11] Tai E, Min A, Rajan DK. A single-center experience with percutaneous interventional management of refractory chylous ascites. Canadian Association of Radiologists Journal. 2020; 846537120929429.

[12] Kim PH, Tsauo J, Shin JH. Lymphangiography with or without embolization for the treatment of post-operative chylous ascites. Annals of Vascular Surgery. 2020; 68: 351–360.

[13] Kawasaki R, Sugimoto K, Fujii M, Miyamoto N, Okada T, Yamaguchi M, *et al.* Therapeutic effectiveness of diagnostic lymphangiography for refractory postoperative chylothorax and chylous ascites: correlation with radiologic findings and preceding medical treatment. American Journal of Roentgenology. 2013; 201: 659–666.

[14] Shimajiri H, Egi H, Yamamoto M, Kochi M, Mukai S, Ohdan H. Laparoscopic management of refractory chylous ascites using fluorescence navigation with indocyanine green: a case report. International Journal of Surgery Case Reports. 2019; 49: 149–152.

[15] Kong TW, Chang SJ, Kim J, Paek J, Kim SH, Won JH, *et al.* Risk factor analysis for massive lymphatic ascites after laparoscopic retroperitonal lymphadenectomy in gynecologic cancers and treatment using intranodal lymphangiography with glue embolization. Journal of Gynecologic Oncology. 2016; 27: e44.

[16] Hur S, Shin JH, Lee IJ, Min SK, Min SI, Ahn S, *et al.* Early experience in the management of postoperative lymphatic leakage using lipiodol lymphangiography and adjunctive glue embolization. Journal of Vascular and Interventional Radiology. 2016; 27: 1177–1186.

[17] Baek Y, Won JH, Kong TW, Paek J, Chang SJ, Ryu HS, Kim J. Lymphatic leak occurring after surgical lymph node dissection: a preliminary study assessing the feasibility and outcome of lymphatic embolization. CardioVascular and Interventional Radiology. 2016; 39: 1728–1735.

[18] Chu HH, Shin JH, Kim JW, Noh SY, Yang WJ, Park S. Lymphangiography and lymphatic embolization for the management of pelvic lymphocele after radical prostatectomy in prostatic cancer. CardioVascular and Interventional Radiology. 2019; 42: 873–879.

[19] Angle JF, Siddiqi NH, Wallace MJ, Kundu S, Stokes L, Wojak JC, *et al.* Quality improvement guidelines for percutaneous transcatheter embolization. Journal of Vascular and Interventional Radiology. 2010; 21: 1479–1486.