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Case Report
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Supermicrosurgical LVA in Abdominal Lymphocele after Gynecologic Cancer Treatment: A Case Report

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Abstract

Introduction: Lymphoceles and lymphorrhea are complications often appearing after lymph node excision in oncologic and transplant surgery. Lymphoceles necessitate treatment in complications such as compression of surrounding structures, infection, or presence of fistulas. Lymphovenous Anastomosis (LVA) has been proven to be effective in the treatment of severe lymphedema, but its application for other indications has been scarce. This case report presents the implementation of an LVA in a patient with recurrent pelvic lymphoceles, transperitoneal and transcutaneous fistulas and consequent lymphorrhea.

Case Presentation: A 64-year-old woman presented with two abdominal lymphoceles following oncologic surgery of a malignant mixed Mullerian tumor of the left ovary and fallopian tube. A subcutaneous (7 cm × 6 cm) and intra-abdominal lymphocele (10 cm × 8 cm) appeared three weeks after surgical treatment and led to pain and grade 3 hydronephrosis of the right kidney. After frustrating repetitive punctures and a revision laparotomy, the patient was assigned to our clinic. The subcutaneous lymphocele could be successfully drained with puncture but the intra-abdominal lymphocele was not accessible for puncture. ICG fluorescent imaging revealed afferent lymphatic vessels conveying fluid to the lymphocele, so they were chosen for LVA implementation. The patient was informed about off-label-use of ICG lymphography. In the follow-up CT two months postoperatively, the intra-abdominal lymphocele completely resoluted and the patient was free of complaints.

Conclusion: The indication of LVA-implementation as a treatment option should be enlarged to further lymphatic complications such as lymphocele and consequent lymphorrhea. Additional studies are needed to confirm its efficacy in more patients.

Keywords: Clinical oncology; Indocyanine green; Lymphatic abnormalities; Lymphatic cysts; Microsurgery

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Introduction

Lymphoceles and lymphorrhea are complications that can occur following lymph node excision in gynecological or urological malignancies, melanoma, or after renal transplantation [1-3]. The incidence of lymphocele after pelvic lymphadenectomy in gynecologic malignancies ranges between 18% [4] and 44% [5]. A lymphocele is a localized collection of lymph. After injury of the lymphatic pathways, a pseudocapsule without endothelial lining develops if the lymph is not absorbed physiologically or drains elsewhere [6]. Lymphoceles are usually asymptomatic however; their removal is indicated in case of compression of surrounding structures, such as blood vessels or nerves. Moreover, there is a risk of infection, fistula formation and impaired wound healing, which are further reasons for surgical intervention. Therapeutic options include interventional radiology procedures (simple aspiration, image guided percutaneous catheter drainage with or without sclerotherapy) and surgical treatment (open or laparoscopic fenestration). Fenestration as marsupialization of the lymph collection into the peritoneal cavity by creating an internal drainage is considered the therapy of choice [7].

Lymphovenous Anastomosis (LVA) is well-established in the treatment of lymphedema of the extremities, but its indication has not been enlarged yet. LVA is a procedure with low invasiveness and is applicable over a wide range of body-areas. Thus, other indications have still to be investigated. Moreover, we intended to test its efficacy in the treatment of recurrent pelvic lymphocele in a patient who had undergone surgical removal of a malignant mixed Mullerian tumor.

Case Presentation

A 64-year-old Caucasian woman presented to our gynecological outpatient clinic with acute abdominal pain and suspected abscess of the lower abdomen. Initially, the patient was referred to us six months before due to a pelvic tumor, which was originally diagnosed as an ovarian tumor. First, she underwent an exploratory laparotomy with hysterectomy, bilateral salpingo-oophorectomy, omentectomy and sigmoid colon resection, after frozen section showed a high-grade adenocarcinoma. The final pathological examination revealed a malignant mixed Mullerian tumor of the left ovary and fallopian tube within filtration of the peritoneum. Besides, pelvic and paraaortal lymphonodectomy was performed. All twenty-one excised lymph nodes were tumor-free. Therefore, the TNM classification was pT3b, pN0 (0/21), cM0, L0, V0. A systemic chemotherapy with carboplatinum and paclitaxel was performed.

Three weeks postoperatively the patient presented with two lymphoceles of the lower abdomen associated with abdominal pain, urinary urgency and swelling of the left lower extremity. Due to a hydronephrosis grade 3 of the right kidney caused by the lymphoceles, a ureteral stent had to be placed and the patient was undertaken repeatedly to ultrasound-guided puncture of the lymphoceles. Finally, four months after the initial surgery and two cycles of chemotherapy, relaparotomy with fenestration of the lymphocele was performed. Three weeks after the second surgery, the patient presented with a recurrence of the lymphocele, which was punctured three more times and another three cycles of systemic chemotherapy were administered.

Physical examination at the date of presentation at our clinic revealed a swelling of the lower abdomen and inflammatory reddening of the lower laparotomy-scar, which was tender and dolorous to palpation. Laboratory tests showed a C-reactive protein level of 26.7 mg/dl (<0.50 mg/dl) and a normal white blood cell count of 6.4×10^9/l (4.0 to 11.0×10^9/l). An abdominal ultrasound examination revealed a large collection of homogeneous encysted fluid of low echogenicity in the subcutaneous tissue of the lower right abdominal wall, as well as a second cystic structure beneath in the lower right abdomen. An initial CT-scan of her abdomen and pelvis confirmed a 10 cm × 8 cm measuring cystic structure in the right pelvis, as well as a new 7 cm × 6 cm measuring cystic structure in the subcutaneous tissue, both with signs of super infection, compression of the bladder and hydronephrosis grade 2 to 3 of the right kidney despite the placed ureteral stent (Figure 1).

Therefore, we diagnosed are current pelvic lymphocele with super infection. Subsequently, the subcutaneous cystic mass was punctured through the abdominal wall under ultrasound control and 400 ml of purulent fluid was drained. Hereafter, only the intra-abdominal lymphocele remained (Figure 2). Culture of the drained intracystic fluid showed *Citrobacter freundii*, *Escherichia coli* and *Klebsiella pneumonie*, all sensitive to Piperacillin/Tazobactam. A systemic antibiotic treatment with Piperacillin/Tazobactam 4 g/0.5 g three times a day was administered for fourteen days post intervention and daily wound irrigations were applied. Here under, the inflammatory parameters decreased well. A follow-up CT-scan of her pelvis showed the intra-abdominal lymphocele smaller with 8 cm × 6 cm in size, but persistent and not accessible for CT-controlled puncture due to a high risk of bowel damage. Therefore, LVA in the area of the right lower extremity was planned.

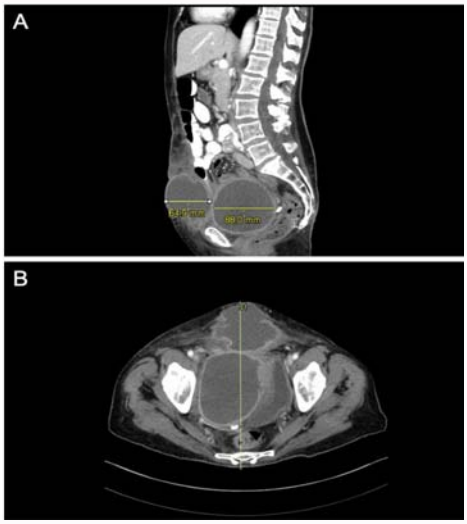

Figure 1: CT of the abdomen and pelvis. A sagittal (A) and transversal image (B) of the pelvic lymphoceles and their measurements are shown.

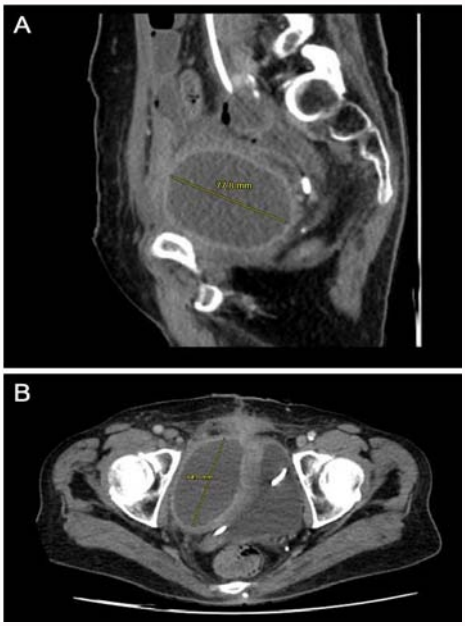

Figure 2: CT scan of the pelvis following puncture of the subcutaneous lymphocele. A transversal (A) and a sagittal image (B) of the remaining intra-abdominal lymphocele is demonstrated.

The preoperative planning was performed using Medtronic EleVision IR Platform system. Indocyanine Green (ICG) 100 L with 0.25% concentration was injected subcutaneously into the first and fourth inter digital space of both feet the day before surgery, identifying the exact fistula localization and locating the lymphatic vessels [8] communicating with the lymphocele (Figure 3). Based on the findings of the ICG lymphography, a precise skin incision could be done and three lymphatic vessels were anastomosed to cutaneous veins under surgical microscopy, using augmented reality and ICG marked navigation (Zeiss Kinevo System).

After LVA, the volume of the right leg and abdomen decreased in size and the secretion via the median laparotomy fistula was minimal. The two-month postoperative control-CT showed the slit-shaped (5 cm in length), clearly decreased lymphocele (Figure 4).

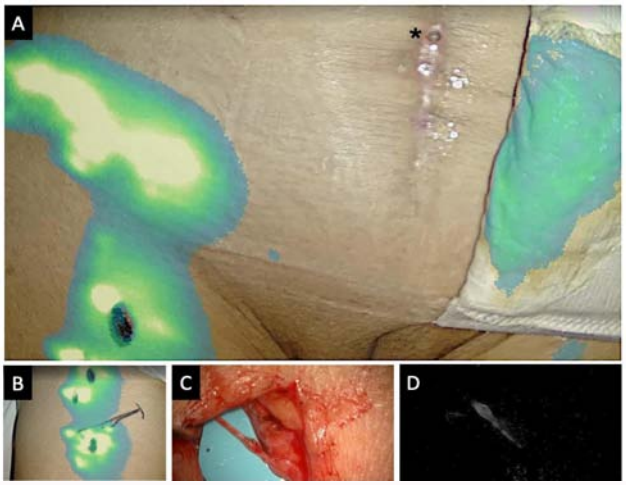

Figure 3: Preoperative ICG- fluorescent lymphography. (A) ICG injection into the first and fourth inter digital space the day before surgery indicated the exact fistula (asterisk) and lymphatic vessel localization. (B) ICG fluorescent imaging identified the lymphatic vessels to connect to the venous system. (C) Lymphovenous end-to-end anastomosis is shown in a native and (D) near infrared image.

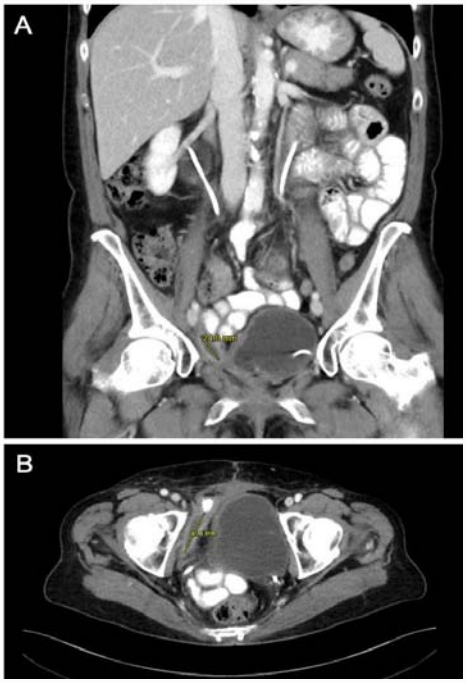

Figure 4: Follow-up CT two months after LVA. The abdominal lymphocele is barely visible and remarkably smaller compared to the previous CT scans in frontal (A) and transversal (B) sections.

Discussion

Lymphatic complications are often inevitable following lymph node dissection in oncologic surgery dissection and adjacent lymphatic vessels are frequently part of the surgical plan [1-3,9]. For post-surgical lymphedema, implementation of LVA has proven to be an effective treatment option, if conservative treatment fails. As to other lymphatic complications such as lymphocele, an adequate surgical treatment with long-lasting results has not been established yet. On the one hand, lymphoceles are frequent complications; on the other hand, they are often asymptomatic [10]. However, if their size leads to compression of surrounding structures such as nerves, vessels or - as in our case - the urinary tract, the patient may complain about symptoms. In these cases, an intervention is indicated. Options like aspiration, drainage or laparoscopic fenestration, frequently need to be performed repeatedly, as lymph collection grows in time [7]. The implementation of LVA, however, represents a permanent solution if the lymphatic vessels conveying fluid to the lymphocele are identified. In this respect, ICG lymphography served as an easily applicable and cost-effective indicator of such vessels.

Our case report describes the medical history of a 64-year-old woman with appearance of two lymphoceles following a complex oncologic intervention including hysterectomy, bilateral salpingo-oophorectomy, omentectomy, sigmoid colon resection, pelvic and paraaortal lymphonodectomy. The size of the lymphoceles led to pain and compression of the right ureter, causing grade 3 hydronephrosis. After frustrating attempts of revision surgery and puncture of the lymphoceles, symptoms persisted. Following consultation in our clinic, the subcutaneous pelvic lymphocele could be punctured and the liquid drained. Culturing of the aspirated fluid revealed bacterial infection. Therefore antibiotic treatment was performed. In contrast to the first lymphocele, the second intra-abdominal lymphocele was not easily accessible to puncture and another therapeutic strategy was necessary. Injection of ICG enabled visualization of lymphatic vessels from the right lower extremity conveying lymphatic fluid to the lymphocele and LVA of these vessels and subcutaneous veins was performed. After two months, a follow-up CT-scan confirmed complete remission of the lymphocele.

Besides vascularized lymph node transfer, LVA is a proven, cost-effective and minimally invasive therapeutic principle in the treatment of lymphedema, alternatively to conservative treatment [11-14]. With regards to further indications for LVA such as its role in lymphocele treatment, only case series and case reports have been published. For instance, Scaglioni et al. [15] recently published a case series confirming LVA efficacy in iatrogenic lymphocele of the thigh. Todokoro et al. [16] described the implementation of LVA in pelvic lymphocele in 2013 already, and in their study, lymphocele completely resolved in six, and partially resolved in five patients.

In the present case report, LVA was proven to be an effective treatment of post-interventional lymphocele, especially after localization of the afferent lymphatic vessels by ICG fluorescent imaging. Indications for implementing LVAs should be enlarged to other lymphatic complications than lymphedema and for further confirmation of their efficacy; interventions on more patients will be required.

Conclusion

Although further case series and prospective studies with longer follow-up periods are needed to confirm its efficacy, LVA appears to be a valuable treatment option for refractory lymphocele and consequent lymphorrhea. ICG represents an advantageous imaging tool in this regard, as it enables to identify afferent lymphatic vessels which need to be connected to respective veins for drainage.

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