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Case Report

Ruptured lymphocele leading to lymphorrhea and wound dehiscence: A treatment approach with drainage and lymphangiography[☆]

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ABSTRACT

A lymphocele is a collection of lymphatic fluid without epithelial lining, commonly occurring as a postsurgical complication. Most lymphoceles do not require treatment, but symptomatic lymphoceles can be successfully treated with surgery, percutaneous drainage and sclerotherapy, or lymphangiography with lymphatic embolization. Although drainage and sclerotherapy are effective in treating lymphoceles, their efficacy decreases in the setting of larger volume, ruptured, or multiseptated lymphoceles. Lymphangiography allows direct visualization of lymphatic vessels and identifies lymphatic leaks and guiding targeted interventions. Moreover, lymphangiography itself has therapeutic potential to directly treat lymphatic leakages. This case report describes a ruptured postkidney-transplant lymphocele, which dehiscence the incision resulting in recalcitrant lymphorrhea. After drainage failed to resolve the lymphocele and sclerotherapy was deemed potentially risky to the kidney transplant, repeated lymphangiography sessions successfully treated the lymphocele and facilitated incisional healing.

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Introduction

A lymphocele is a collection of lymphatic fluid with no epithelial lining, most frequently iatrogenic, arising after surgical intervention in lymphatic rich regions. Lymphoceles are generally asymptomatic but can cause mass effect and compress surrounding structures such as ureters, veins, or nerves causing hydronephrosis, edema, or pain, respectively [1–2]. Symptomatic lymphoceles are treated either surgically or with image-guided interventions. Current image-guided lymphocele management includes percutaneous aspiration, drain placement, sclerotherapy, lymphangiography, lymphatic embolization, and lymphaticovenular anastomosis [1–5].

Intranodal lymphangiography is a diagnostic and therapeutic procedure used to identify and treat lymphatic leakage. While lymphangiography is commonly performed to guide embolization, repeated lymphangiography alone can be therapeutic due to the properties inherent to ethiodized oil [1,6]. Therapeutic lymphangiography has been shown to be an effective treatment for various lymphatic leakages, including lymphoceles, with success rates heavily correlated to the preintervention volume of lymphatic drainage per day [6–9]. This case highlights the successful use of percutaneous drainage and repeated lymphangiography to treat a ruptured postrenal transplant lymphocele which presented with lymphorrhea, wound dehiscence, and lower extremity edema.

Case presentation

A 63-year-old male, approximately 1.5 months postrenal transplant, presented to the emergency department with suspected renal transplant dysfunction and bilateral lower extremity pitting edema. The transplant incision site was healing poorly with light yellow, clear fluid leaking through the wound. Transplant renal ultrasonography (US) and subsequent computed tomography (CT) revealed a multilobu-

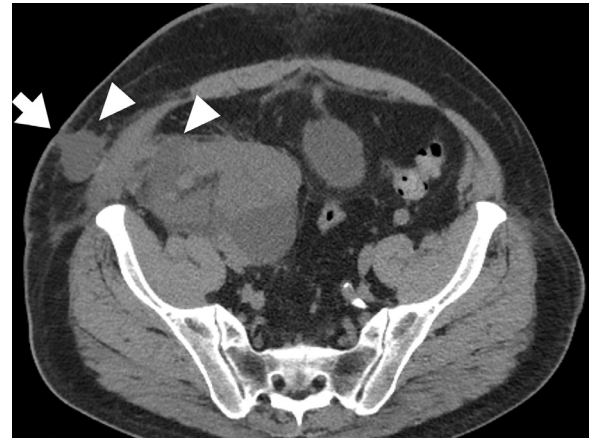


Fig. 1 – Axial CT image of the pelvis reveals a right lower quadrant renal transplant surrounded by fluid collections extending to the subcutaneous tissue (white arrowheads). The skin is discontinuous and the site of incision dehiscence and fluid leakage (white arrow).

lated perinephric collection of simple fluid density measuring $6.6 \times 5.7 \times 11.8$ cm (Fig. 1). Under US guidance, transplant kidney biopsy was performed to evaluate for rejection in standard fashion. This was followed by US- and CT-guided placement of an 8 French drainage catheter (Uresil LLC, Skokie, IL, USA) into the perinephric collection, which was maintained on bulb suction. In total, 140 mL of clear, yellow fluid was aspirated and sent for cell count, creatinine, triglyceride, and microbiology. Despite drain placement, clear, yellow fluid continued leaking from the incision, which was partially dehiscid (Fig. 2).

Fluid testing revealed no neutrophils or red blood cells, but a high number of lymphocytes (99%), as well as negative microbiology, and low creatinine (3.7 mg/dL) and triglyceride (< 20 mg/dL) values. Ten days later, CT imaging of the drain revealed a persistent lymphocele in the right lower quadrant and the drain within subcutaneous tissue outside



Fig. 2 – Photograph of the right lower quadrant dehiscid incision with clear, yellow fluid continuously leaking despite drain placement.

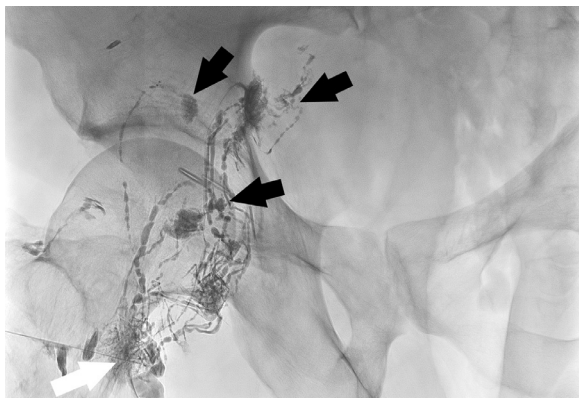


Fig. 3 – Right inguinal nodal lymphangiography was performed with a 25-gauge needle directly injecting contrast through a lymph node (white arrow). Multiple foci of lymphatic leakage and contrast pooling are present in the area of the lymphocele and surgical clips (black arrows).

of the collection. Therefore, drain check and lymphangiography was performed. Under US guidance, a 25-gauge chiba needle was positioned within a right inguinal lymph node. Intra-nodal lymphangiography with Lipiodol (Guerbet, Princeton, NJ) revealed multifocal lymphatic leaks into the lymphocele (Fig. 3). At that time, nodal lymphatic embolization was deferred given the potential of worsening 3+ pitting edema in the right lower extremity. The drain was repositioned within the lymphocele, and contrast injection was performed in preparation for potential sclerotherapy, which subsequently led to contrast leakage at the incision site. Therefore, sclerotherapy was aborted to avoid further injury to the dehiscid incision.

Approximately 4 weeks following initial lymphangiography, repeat drain check again revealed continued leakage from the incision and into the adjacent tissues (Fig. 4). So, sclerotherapy was again deferred. Repeat right intra-nodal lymphangiography demonstrated persistent lymphatic leaks adjacent to the lymphocele. Lymphatic embolization was again deferred to avoid the potential of worsening of the right lower extremity edema. Approximately 4 weeks later, following healing of the surgical incision, cessation of the drain output, and resolution of lower extremity edema, the drainage catheter was removed (Fig. 5). At 6-month follow-up, the incision remained healed, lower extremity edema had resolved, and the renal transplant continued to function normally.

Discussion

This case highlights the successful use of a drainage catheter and repeated lymphangiography to resolve a multiloculated lymphocele, which was associated with lower extremity edema and incision dehiscence resulting in lymphorrhea, in a postkidney transplant patient. Standard treatment approaches for lymphocele include drainage with sclerotherapy or lymphangiography with lymphatic embolization [1,7,9,14].

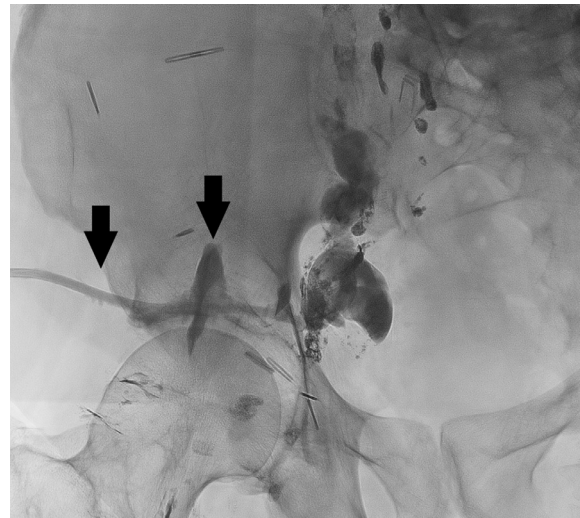


Fig. 4 – Fluoroscopic image of contrast injection into the right lower quadrant lymphocele drain revealed contrast refluxing along the drain track, into adjacent soft tissue, and leaking out outside of the body (black arrows).

However, sclerotherapy is contraindicated in cases of multilocular lymphocele rupture and lymphorrhea, and lymphatic embolization may not be suitable for patients with worsening lower extremity edema. Two sessions of intra-nodal lymphangiography demonstrated areas of lymphatic vessel injury with leakage of ethiodized oil into the lymphocele. In combination with drainage, lymphangiography reduced the size of the lymphocele, promoted wound healing, and resolved the lower extremity edema. This case redemonstrates the application of lymphangiography as an effective treatment option for complex lymphocele and other lymphatic leakages. Notable indications for lymphangiography, in this case, were multifocal nature of the lymphatic leakage, lymphorrhea, wound dehiscence and lower extremity edema.

Diagnosis and management of lymphoceles since their initial description in the 1950s has evolved greatly with advancements in imaging and percutaneous therapies [10]. Similarly, lymphangiography was first described through pedal access in the 1950s [11]. However, the intersection of routinely using lymphangiography to diagnose and treat lymphocele would not occur until after the advent and widespread adoption of intranodal lymphangiography as an easier, less time-consuming alternative to visualize lymphatic flow. Intranodal lymphangiography is currently considered the gold standard for lymphatic system diagnostic imaging [12].

Lymphangiography has since become an invaluable tool for guiding direct interventions for chylothorax, chylous ascites, lymphatic fistulas, lymphocele, and lymphorrhea [13–16]. Repeated lymphangiography in combination with drainage is a therapeutic alternative for lymphoceles with poor candidacy for sclerotherapy and embolization. The therapeutic mechanism of ethiodized oil, which is used in lymphangiography, remains largely unknown with 2 suggested processes leading to embolization: accumulation within leaks inducing inflammation and saturation of lymphatic vessels causing lymph redi-

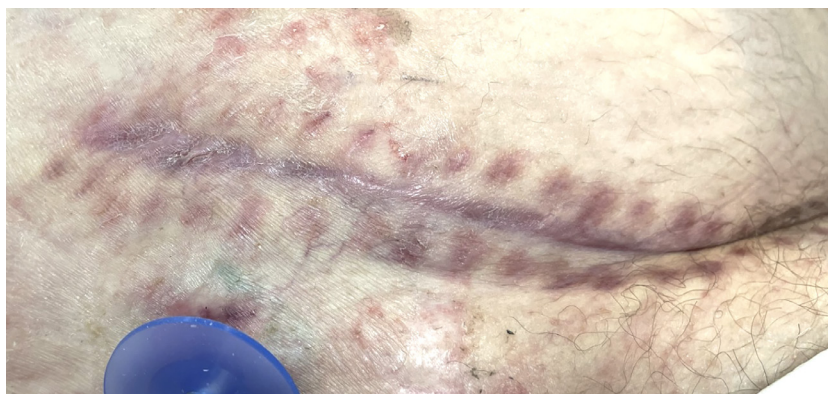


Fig. 5 – Photograph of the right lower quadrant healed incision. The drain was subsequently removed.

rection away from leaks [5]. Nonetheless, lymphangiography with ethiodized oil remains as an effective treatment option for many lymphatic leakages.

Potential disadvantages of lymphocele management with lymphangiography may include multiple procedures, cost, and lower efficacy for large, high-volume leaks. However, percutaneous sclerotherapy and/or lymphatic embolization may address these disadvantages in isolated cases and the therapeutic utility of lymphangiography alone should not be overlooked. For appropriately selected patients who may have contraindications for other therapies, lymphangiography should be considered as a safe and well-tolerated treatment option for lymphatic leakages.

In conclusion, continuous drainage and repeat lymphangiography could be effectively utilized for treatment of complex lymphocele and other lymphatic leakages or in patients with lower extremity edema.

Patient consent

IRB approval was granted for this publication. Written informed consent was obtained from the patient.

Author contributions

BSM: Consultant and Speaker: Guerbet; Scientific Advisory Board: Balt Medical. NN: Advisory Board: Boston Scientific and CAPS Medical; Consultant and Speaker: Boston Scientific; Safety Monitoring: Microbot Medical.

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