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Case Report
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Lipiodol-based Lymphangiography and Glue-based Embolization of Retroperitoneal Lymphatic Vessels to Treat Symptomatic Retroperitoneal Lymphocele: A Case Report

*Nariman Nezami, Haddy Jarmakani, Angelo Marino, Todd Schlachter, Igor Latich, Rajasekhara Ayyagari and Juan Carlos Perez Lozada**

Department of Radiology and Biomedical Imaging, Yale School of Medicine, USA

Abstract

Lymphocele occur due to the disruption of lymphatic vessels typically in the setting of trauma or surgery. A number of procedures have been introduced to treat this entity, but none have been entirely successful. Here, we present a 67-year-old female diagnosed with stage III ovarian cancer who developed a retroperitoneal lymphocele after robotic hysterectomy, bilateral salpingo-oophorectomy, and tumor debulking. She had multiple unsuccessful sessions of sclerotherapy over two months. Eventually, the lymphatic vessels supplying the lymphocele were mapped by lipiodol injection in the inguinal lymph nodes and embolized withN-butyl-2-cyanoacrylate *glue*. This resulted in resolution of the lymphocele and symptoms. Lipiodol-based mapping and glue embolization of retroperitoneal lymphatic vessels is a safe and feasible procedure to treat retroperitoneal lymphoceles.

Keywords: Lymphocele; Retroperitoneal; Embolization; *Glue*; Lymph vessel; Lipiodol

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Correspondence:
Juan Carlos Perez Lozada, Department of Radiology and Biomedical Imaging, Yale School of Medicine, USA, Tel: +1 (203) 400-7650; Fax: +1 (203) 785-7026;
E-mail: juancarlos.perezlozada@yale.edu

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Introduction

Lymphocele is a known rare complication of surgery or consequence of trauma, caused by disruption of lymphatic vessels. It mainly occurs after renal transplantation, vascular, spinal, urologic or gynecologic surgeries [1-6]. Most patients are either diagnosed incidentally on post-surgical follow up imaging or present with secondary symptoms due to mass effect. Although surgical resection was initially considered the gold standard treatment for lymphocele [7], its association with high rate of recurrence, infection, and need for hospitalization quickly replaced the open procedure with laparoscopic approach [8-10].

In recent years, minimally invasive procedures ranging from simple aspiration, catheter-based drainage, and transcatheter sclerotherapy have become popular. Simple aspiration alone is frequently inadequate [8,11]. However, a combination of catheter-based drainage and transcatheter sclerotherapy produce results comparable to surgery [12]. Sclerotherapy causes wall irritation by inducing local inflammation and fibrosis of the lymphatic channels, thus obliterating the lymphatic leak [11]. While there is abundant literature on surgical and trans-catheter approaches, there are only a few reported cases of glue embolization of pelvic lymphocele. Our case demonstrates a post pelvic surgery retroperitoneal lymphocele treated with lipiodol mapping and glue embolization of the feeding lymphatic vessels.

Case Presentation

A 67-year-old female with past medical history of recently diagnosed stage III papillary ovarian cancer, who is status post adjuvant chemotherapy and robotic assisted hysterectomy, bilateral salpingo-oophorectomy, tumor debulking with Omentectomy. A month after surgery, ultrasonography of the retro peritoneum performed for left sided pelvic/back pain showed new moderate hydronephrosis of the left kidney without visualization of the left ureterovesicaljet, while free fluid within the pelvis appeared less prominent compared to the ultrasound done prior surgery (Figure 1). Contrast-enhanced CT scan of abdomen and pelvis five days later revealed significant left-sided hydroureteronephrosis involving the proximal third of the left ureter, secondary to mass effect from a large 5.6 cm cystic retroperitoneal hypoattenuating collection, most consistent with a retroperitoneal lymphocele (Figure 2).

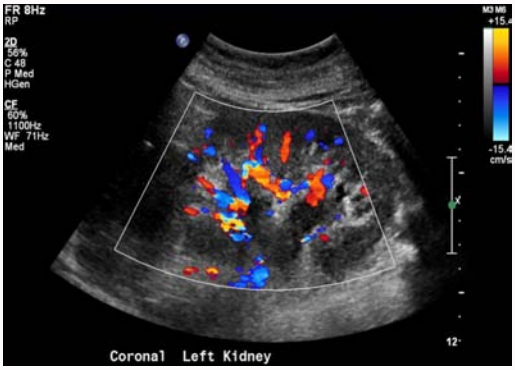

Figure 1: Color Doppler ultrasound using a 5 MHz curved transducer shows left renal hydronephrosis appreciated by renal pelvic dilatation.

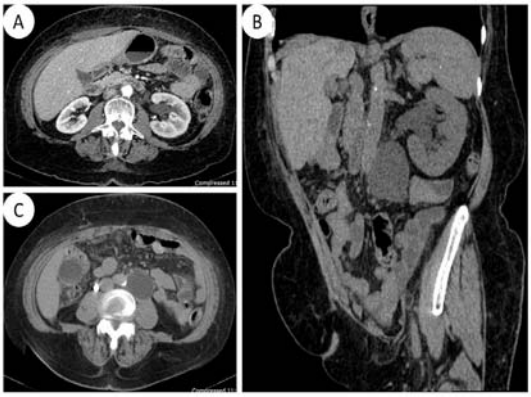

Figure 2: A. Transverse view of contrast CT scan show shydronephrosis and dilatation of the collecting system to the level of the proximal ureter. **B.** Coronal view of contrast CT scan demonstrates a 3.6 cm x 4.1 cm x 5.6 cm hypodense retroperitoneal collection. **C.** On the transverse view of contrast CT scan, the left ureter is not distinguishable from fluid collection and the left ureter is not visualized distal to this cystic fluid collection.

Subsequently, the patient underwent cystourethroscopy which showed a normal urinary bladder with moderate hydroureteronephrosis is caused by obstructive retroperitoneal lymphocele. A double-J stent was then placed to alleviate obstruction. Follow-up CT guided aspiration of the retroperitoneal collection yielded 35 mL of clear yellow fluid, which was negative for infection on culture and sensitivity tests. A CT scans of the abdomen one and half months later showed a persistent/recurrent retroperitoneal fluid collection (Figure 3). This, the decision was made to carry out CT-guided percutaneous left retroperitoneal lymphocele aspiration and drainage catheter placement. Twenty-five mL of clear yellow fluid was aspirated, an 8 French pigtail drainage catheter was placed (Figure 4), and sclerotherapy performed using 10 mL of 98% (absolute) ethanol which was left to dwell for one hour prior to drainage. However, the lymphocele persisted after five sessions of sclerotherapy with absolute alcohol. Consequently, the patient underwent left inguinal lymphangiography and glue embolization of the left retroperitoneal/iliac supplying lymphatic vessels to treat lymphocele.

Technique

First, the hilum of a left groin lymph node was accessed with a 9 cm 25-gauge spinal needle under ultrasound guidance (Figure 5A). Lipiodol was then slowly infused and observed to pass into the left inguinal lymphatic chains (Figure 5B). After about 30 mins, two

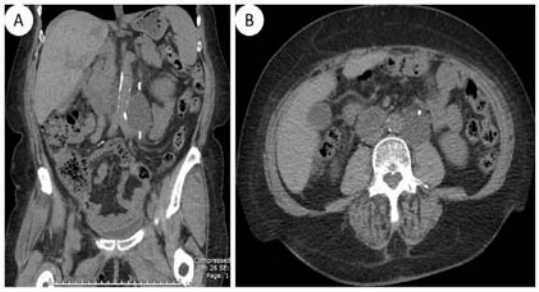

Figure 3A and 3B: A new left ureteral stent is seen in place with no residual hydronephrosis. The both coronal and transverse views demonstrate persistent left retroperitoneal fluid collection and a new left ureteral stent, revealing independent nature of the cystic fluid collection from the left ureter. These findings also explain left proximal hydroureteronephrosis as a consequence of mass effect from this fluid collection.

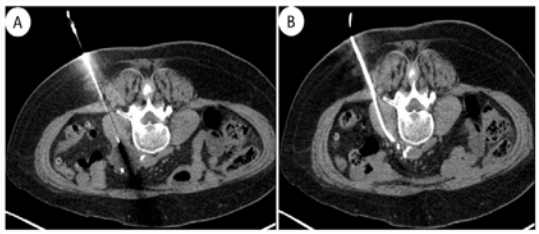

Figure 4A and 4B: These transverse views show CT-guided percutaneous left retroperitoneal lymphocele aspiration and drainage catheter placement through the left psoas muscle.

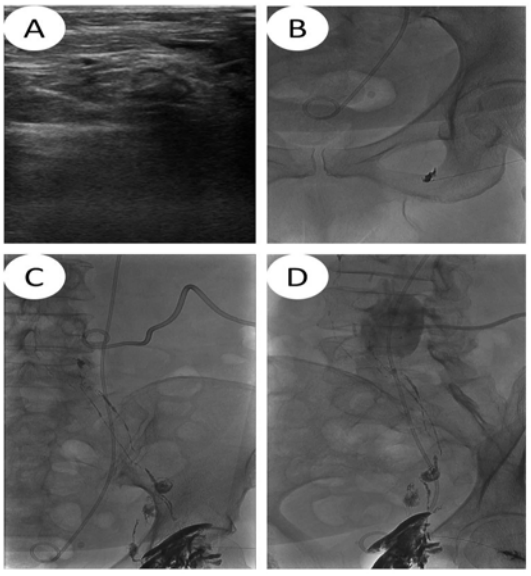

Figure 5: A. A left groin lymph node was accessed with a 9 cm 25-gauge spinal needle under ultrasound guidance. **B.** Fluoroscopy image showing intranodal lipiodol injection. **C.** Extravasation of lipiodol into the soft tissues is shown. This was corrected by puncturing an adjacent left inguinal lymph node. Lipiodol seen passing into the left iliac lymphatic vessels. After about 30 minutes, two retroperitoneal/iliac lymphatic vessels are progressively opacified over the mid left retroperitoneal space, caudal to the retroperitoneal lymphocele. **D.** Contrast injection into the cavity via indwelling catheter identifies left lateral lymphatic vessel as a contributor to the persistent/recurrent left retroperitoneal lymphocele.

iliac lymphatic vessels were progressively opacified over the mid left retroperitoneal space to the level of retroperitoneal lymphocele (Figure 5C). Upon injection of contrast into the left retroperitoneal

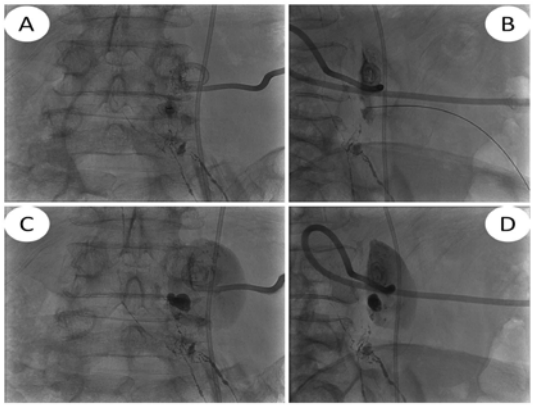

Figure 6: A. Anteroposterior and lateral views. A 22-gauge Chiba needle was transabdominally advanced with straight approach toward the visualized mid left retroperitoneal/iliac lymphatic vessel just caudal to the lymphocele. Digital lymphangiogram performed through the needle shows a single retroperitoneal/iliac lymphatic vessel, with contrast extravasation into the left retroperitoneal lymphocele. **B.** Embolization with 1 ml of n-BCA glue in a 1:1 ratio with lipiodol as well as tantalum powder for improved visualization. Glue distributing evenly within the left lateral retroperitoneal/iliac lymphatic vessel caudal to the lymphocele.

cavity, the left lateral retroperitoneal lymphatic vessel was identified as a potential supplying vessel to the lymphocele (Figure 5D).

The left lateral retroperitoneal/iliac lymphatic vessel, at the level of L4-L5 and distal to the lymphocele, was successfully accessed via a 22-gauge Chiba needle through the abdomen under direct fluoroscopic visualization (Figure 6A). Digital lymphangiogram demonstrated contrast extravasation into the left retroperitoneal lymphocele. Next, embolization was performed with 1 ml of N-butyl-2-cyanoacrylate *glue* (n-BCA) glue mixed in a 1:1 ratio with lipiodol along with tantalum powder for improved visualization (Figure 6B). The glue was seen to distribute evenly within the left retroperitoneal lateral lymphatic vessel distal to the lymphocele. Contents of the lymphocele were then aspirated through the left upper back drainage catheter, confirming the collapse of the left retro peritoneum lymphocele. Opacified glue was demonstrated to reach the inferior aspect of the cavity. Follow-up drainage catheter check five days later showed no further output. The catheter was thus removed.

Discussion

There are well described techniques in the surgical and interventional literature regarding treatment of refractory lymphoceles, the most common of which are surgical resection, aspiration/drainage, and sclerotherapy [1-5]. However, these interventions have been associated not only with added risk to the patient but high treatment failure rates [1-6]. We present a case of post-surgical retroperitoneal lymphocele that was successfully treated by lymphatic vessel mapping and glue embolization after five unsuccessful sessions of sclerotherapy. Sclerotherapy and drainage are commonly utilized as first line treatment. Chemical sclerotherapy is usually performed by first aspirating the lymphocele cavity with subsequent infusion of sclerosant, often requiring repeated sessions. Commonly used sclerosants include alcohol, povidone iodine, and doxycycline [12]. Success of sclerotherapy has been found to be directly related to the size of the lymphocele cavity, with larger cavities more likely to be resistant to treatment. Complications of sclerotherapy include catheter related infection, potential allergy or adverse reaction to sclerosant, and spillage of sclerosant into the retro peritoneum causing massive inflammation [13]. Glue embolization on the other hand is an outpatient procedure requiring only one visit, which eliminates the potential complications from various sclerosants [14].

With larger lymphoceles or failed sclerotherapy and drainage, traditional surgical options include laparoscopic surgery, open marsupialization, and percutaneous imaging guided lymphatic ligation. These surgical interventions, while typically successful, are associated with longer hospital stay and increased infection rates. Additionally, patients must be able to tolerate further surgery, making this option unavailable for more complicated patients [15]. Conversely, glue embolization is a two-step procedure during which one identifies the leaking lymphatic vessel and proceeds with embolization all in a single session [16].

This renders lymphatic mapping by ultrasound guided intranodal lymphangiogram and glue embolization a viable alternative to the established approaches, minimizing the risk of potential complications and decreasing the length of treatment commonly associated with surgery and sclerotherapy [17]. Access to the retroperitoneal lymphocele can be performed with cone beam CT, temporary catheter placement and subsequent lymphangiogram and glue embolization with fluoroscopic imaging as a single procedure. Current noninvasive methods aim treatment at collapsing the walls of the lymphocele, while a few treatment options target the feeding lymphatic pathway directly. This is where a minimally invasive technique such as lymphography and selective glue embolization of feeding lymphatics may be invaluable as a stand-alone treatment option, or in conjunction with drainage and sclerotherapy [18], as was utilized in our case.

This method of selective glue embolization of lymphatic leaks is well described with thoracic duct lymphangiography for post-surgical leaks [19,20]. To our knowledge, there is sparse evidence of the application of this technique elsewhere. In the case of our patient, a similar technique was applied to the inguinal lymphatic pathway involving the retro peritoneum. Although lymphangiography and embolization of the supplying lymph vessels was performed after sclerotherapy failure, it has the potential to be the first-line treatment. Main advantage of this technique when compared to sclerotherapy that it requires only one visit. Minimally invasive nature of the procedure renders it a very attractive option in comparison to surgery or sclerotherapy.

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