Case Report

DOI: 10.7860/JCDR/2022/58956.17151

Ethiodised Oil Lymphangiography in the Management of Chyluria

Radiology Section

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ABSTRACT

Chyluria is clinical condition in which there is excretion of chyle in urine. The most common cause of chyluria is lymphatic filariasis. This case report describes imaging findings of non filarial chyluria due to retroperitoneal lymphatic cyst and its successful management by minimally invasive percutaneous lymphangiography procedure. The patient presented with complaints of passing milky white urine which was confirmed to be chyluria. After a non successful trial of conservative approach in form of high-protein and low-fat diet, percutaneous ethiodised oil lymphangiography was performed, which resulted in immediate symptomatic improvement and no recurrence on follow-up.

Keywords: Chyle, Chylomicrons, Interventional radiology, Lymphatic cyst, Sclerotherapy

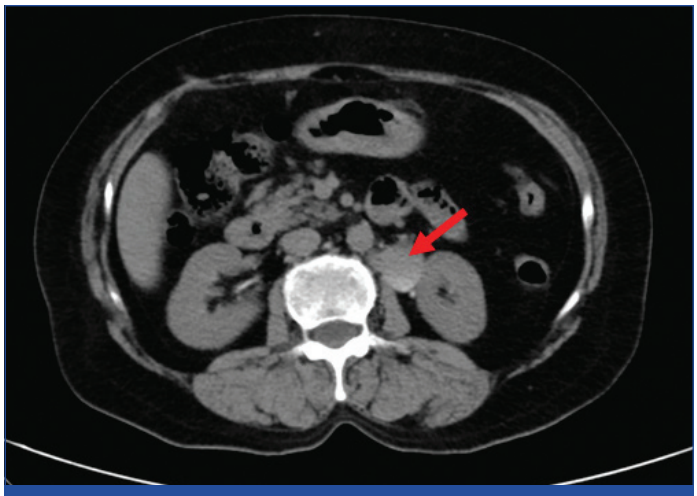
CASE REPORT

A 67-year-old female presented to the Department of Interventional Radiology with complaints of intermittent left flank pain and persistent milky white urine, since one year. She had no co-morbidities, no surgical history, and no visit to filarial endemic areas.

The woman weighed 56 kg, with a body temperature of 37°C, pulse rate of 76/min, and blood pressure of 110/70 mmHg. The heart and lung examinations were normal. The abdomen was soft with non palpable liver and spleen. There was no shifting dullness.

Urine analysis revealed sterile urine, massive proteinuria (2g/24 hours), and elevated triglyceride levels (600 mg/dL) in post prandial urine samples, thus diagnosing Chyluria. As per protocol, she was advised conservative management in form of low-fat, high-protein diet. However, even after five months follow-up, she did not had any symptomatic improvement.

As part of further investigations, Computed Tomography (CT) scan was done which revealed a well-defined spherical retroperitoneal cystic lesion of approximate size 22×23×23 mm (CC×AP×TT) in left para-aortic region, medial to the lower pole of left kidney likely Cystic Lymphangiectasia [Table/Fig-1]. The possible cause for chyluria was fistulous communication between this cyst and left renal pelvis or proximal left ureter.



[Table/Fig-1]: Axial post contrast CT scan image showing the enhancement in left sided retroperitoneal lymphatic cyst.

As part of definitive treatment, minimally invasive percutaneous lymphangiography was planned in the Cathlab, under fluoroscopy guidance. Under ultrasound guidance and local anaesthesia (5 mL of 2% Lignocaine), three inguinal nodes on either side were punctured with 22 G needle. With the needle tip at the junction of cortex and medulla of each lymph node, five mL ethiodised oil (Lipiodol, Guerbet) was slowly injected under fluoroscopy guidance on either side, to visualise it ascend in the lymphatic channels [Table/Fig-2]. and prevent its entry into veins. Approximately, 40 minutes post completion of the injection, there was intense opacification of the left sided lymphatic cyst [Table/Fig-3,4], and immediate non contrast CT scan of the pelvis revealed ethiodised oil inside the lumen of urinary bladder [Table/Fig-5], confirming the presence of fistulous communication. As the definitive site of fistulous communication was not demonstrated, it was decided to stop at this stage.

The patient was shifted to Intensive Care Unit (ICU) post procedure for close monitoring. Post 24 hours of procedure, the urine became completely normal in appearance. Microscopic urine examination revealed resolution of proteinuria and absence of triglycerides in urine. After discharge, the patient was followed after two weeks, one, three and six months with no recurrence of chyluria.

[Table/Fig-2]: Fluoroscopy image after percutaneous lymphangiography showing presence of ethiodised oil in pelvic lymphatics.

[Table/Fig-3]: Fluoroscopy image after percutaneous lymphangiography showing presence of ethiodised oil in left sided retroperitoneal lymphatic cyst.

[Table/Fig-4]: Axial non-contrast CT scan image showing the dense deposition of ethiodised oil in the left sided retroperitoneal lymphatic cyst post minimally invasive lymphangiography done via inguinal lymph nodal injection.

[Table/Fig-5]: Axial non-contrast CT scan image (done immediately after lymphangiography) showing the presence of ethiodised oil inside lumen of urinary bladder suggesting presence of abnormal communication between the retroperitoneal lymphatics and urinary system.

DISCUSSION

Chyle is a combination of proteins, emulsified fat and fibrin. It is formed by lymph and absorbed dietary fats which are converted to chylomicrons. It is transported by the lymphatic channels to thoracic duct which opens into the left subclavian vein. Chyluria is classified as parasitic and non parasitic [1]. In endemic areas, *Wuchereria bancrofti* is considered to be the parasitic cause for Chyluria [2]. Tuberculosis, congenital anomalies, trauma, pregnancy, abscess, postsurgery, infections and malignancy are the common causes of non parasitic Chyluria [2]. Chyluria occurs when there is formation of abnormal communication between the lymphatic vessels and urinary tract resulting in milky white urine. The site of fistulous communication can be at the renal pelvis, ureter, urinary bladder or the prostatic urethra [2]. The patients of chyluria may present with renal colic due to the passage of milky-white urine along with clots, dysuria, haematuria and urinary tract infections. In some severe forms, weight loss, cachexia, malnutrition, hypoproteinaemia and immunosuppression can be seen [2].

Majority of the patients present with milky white colour of urine [3]. Chyluria due to parasitic cause presents with concomitant genital manifestations, lymphatic obstruction in limbs, cellulitis, abscesses and haematuria [4].

On macroscopic examination, chyluria has milky appearance which may be sometimes mixed with fibrin and blood clots. A postprandial urine sample is usually recommended for investigations [2]. The milky white urine turns clear on addition of fat solvent such as ether. When Sudan III stain is added, there is superficial layer of red-stained fatty particles lying on clear urine after resting. Estimation of urinary chylomicrons is the most specific and sensitive test for chyluria [5]. Triglyceride levels above levels 15 mg/dL are indicative of chyluria. Filarial antigen detection in the urine and serum can be done in suspected parasitic causes [6].

Conservative management is successful in upto 70% cases of chyluria [7]. High fluid intake, fat restriction with addition of green leafy vegetables and multivitamins is recommended. In some severe intractable cases, total parenteral nutrition has been proposed. [8]. There is very limited information on recommended duration of conservative management in literature, however, increasing severity of symptoms and signs of nutritional deficiencies warrant additional treatment options. In filarial patients, dietary modifications along with medications are needed. The recurrence rates after conservative management have been as high as 80% [9].

The minimally invasive therapy includes sclerotherapy, ethiodised oil lymphangiography or endoscopic coagulation. 0.1-3% silver nitrate, 0.2% povidone iodine, 1-25% sodium iodide, 10-25% potassium bromide, 50% dextrose and hypertonic saline are the various sclerosing agents utilised [3]. In this sclerotherapy procedure, under optimal anaesthesia, 5F ureteric catheter is passed into the renal pelvis and sclerosant injected at site of fistulous communication which reaches the adjacent lymphatics via fistulous communication [8]. It induces oedema and inflammation which results in subsequent fibrosis for long lasting symptomatic relief [6]. However, this usually requires multiple treatment sessions over 6-8 weeks [6]. The minor and self-limiting complications post sclerotherapy include nausea, flank pain and haematuria [10]. More serious conditions like renal failure, anuria, pelvicalyceal cast formation and acute necrotising ureteritis have been also reported [11]. The recurrence rates post sclerotherapy have been reported to be 13-41% [12].

Image-guided lymphatic interventions utilise imaging modalities to provide minimally invasive targeted therapies to block the abnormal lymphatic connections, which was previously performed by open surgery [13-15]. Recently published reports describe diagnostic and therapeutic role of lymphangiography for lymphatic leakage [16,17]. The ethiodised oil undergoes inflammatory and granulomatous reaction during its extravasation leading to its therapeutic effect along with its embolic properties due to high viscosity [17]. For cases of massive postoperative chylothorax, thoracic duct embolisation is minimally invasive procedure alternative to surgical ligation of thoracic duct [18]. The lymphatic system is opacified by lymphangiography and then the cistern chyle or thoracic duct is

catheterised through transabdominal approach and subsequently embolised. When the thoracic duct cannulation is not successful, thoracic duct obliteration of cistern chyle obliteration can be done. Needle perforation of the proximal thoracic duct/cistern chyle is done within the abdomen which prevents ascend of lymph which leaks into the peritoneal cavity and is absorbed by visceral organs.[18]. The complications of ethiodised lymphangiography include intra-alveolar haemorrhage, oil embolism to pulmonary vasculature, allergic reactions and oil extravasation in soft tissue at site of injection [17]. The contraindications for the procedure are pulmonary insufficiency (can be exaggerated due to pulmonary embolism) and right to left cardiac shunt (risk of cerebral embolism). The overall complication rates are low when volume of ethiodised oil used is less than 10 mL.

When conservative and minimally invasive treatment options fail, invasive surgical treatments are considered. The various operative techniques described are chylolymphatic disconnection and creation of lymphovenous anastomoses, omental wrapping and auto-transplantation or nephrectomy [19]. The overall success rate above 98% has been described for chylolymphatic disconnection [1]. Simple nephrectomy is done in cases of non functioning kidney after conventional lymphovenous disconnection [6]. Lympho-venous anastomosis results in lymphatic diversion and reducing intralymphatic pressures and is more physiological [20]. With the introduction of advanced laparoscopic and robotic techniques, the complication rates have significantly reduced [21].

CONCLUSION(S)

Lymphangiography is a relatively safe, minimally invasive, and reliable interventional modality which has diagnostic and therapeutic role in cases of lymphatic leakages. The need for morbid surgery is avoided.

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PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Jul 09, 2022
- Manual Googling: Sep 10, 2022
- iThenticate Software: Oct 03, 2022 (11%)

ETYMOLOGY: Author Origin

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Jul 08, 2022**
Date of Peer Review: **Sep 13, 2022**
Date of Acceptance: **Oct 04, 2022**
Date of Publishing: **Nov 01, 2022**