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Case Report

Urology Section

Successful Iohexol Sclerotherapy for Non-parasitic Chyluria: A Case Report

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ABSTRACT

Chyluria is a medical condition characterised by the passing of milky urine due to the abnormal drainage of lymphatic fluid, known as chyle, into the urinary tract. This condition can result from a variety of aetiologies, both parasitic and non-parasitic, with Wuchereria bancrofti being the primary causative agent. Diagnosis includes clinical presentation, laboratory investigations, and imaging studies. Management encompasses a wide array of conservative therapies and surgical interventions. A 60-year-old male presented with chyluria and chyle clots for one year. He had received medical treatment for filariasis and dietary modifications at various hospitals before being referred to this centre. The patient was investigated and diagnosed with non-parasitic chyluria due to a left pyelo-lymphatic fistula and subsequently underwent sclerotherapy with Iohexol contrast, which resulted in complete resolution. This minimally invasive procedure involves the injection of a contrast agent that helps seal affected lymphatic vessels, preventing further chyle leakage into the urinary system. Severe chyluria can be effectively managed with Iohexol contrast sclerotherapy. This case underscores the importance of timely intervention and the use of sclerotherapy with Iohexol contrast as a treatment for severe chyluria, offering a promising therapeutic option for patients with persistent or complicated cases.

Keywords: Infectious diseases, Pyelo-lymphatic fistula, Urology

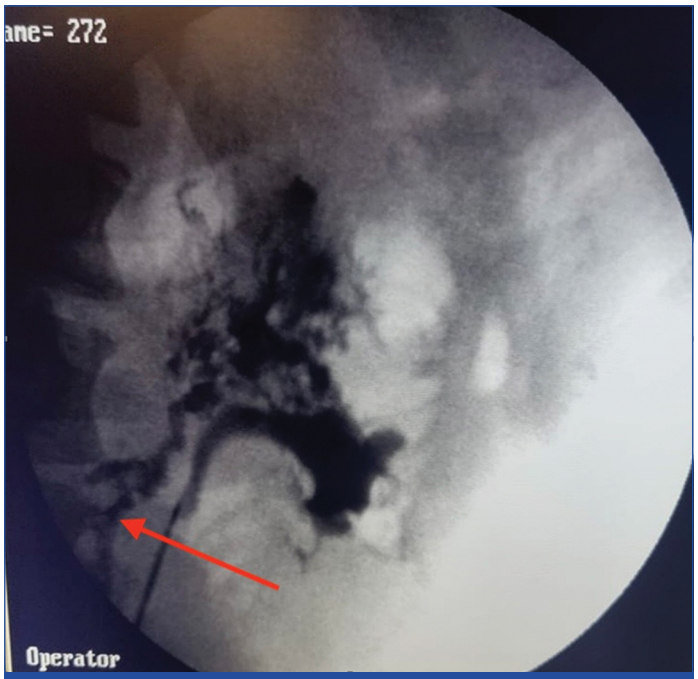
CASE REPORT

A 60-year-old Indian male from Maharashtra state presented to the urology department with complaints of passing milky white urine for one year, associated with occasional passage of thick white clots. This was accompanied by irritative lower urinary tract symptoms and occasional colicky pain in the left flank. The patient reported significant weight loss over the past six months. There was no history of haematuria, burning micturition, or fever. No significant medical history, comorbidities, or past surgeries were reported. The patient had previously received anti-filarial medications, such as Hetrazan, and dietary modifications that included a low-fat, high-protein diet with Medium Chain Triglycerides (MCT) supplementation at other hospitals for over a year.

On examination, the patient was haemodynamically stable, and his systemic examination was within normal limits. The urine appeared milky, with a protein level of +, while the remainder of the analysis was normal. A 24-hour urine test for albumin was normal, the fasting lipid profile was normal, and serum albumin was measured at 3.5 mg/dL. The haemogram, liver, and renal function tests were unremarkable. A peripheral blood smear for filariasis returned negative results. An abdominal ultrasound and chest X-ray were conducted, both of which were normal. A urine culture was sterile. The provisional diagnosis for this case was chyluria, with differential diagnoses of phosphaturia and bacterial urinary tract infection being considered.

The patient was planned for cystoscopy and a bilateral Retrograde Pyelogram (RGP) with sclerotherapy. After obtaining written and informed consent, the patient underwent cystoscopy, which revealed milky white urine coming from the left ureteric orifice. The left RGP [Table/Fig-1], performed through a 5Fr ureteric catheter, demonstrated a left pyelo-lymphatic fistula with dye entering the lymphatic system. Ten millilitres of undiluted Iohexol contrast was used for sclerotherapy; the solution was injected slowly under fluoroscopic guidance, held for three minutes, and then released. A single sitting of Iohexol sclerotherapy successfully alleviated the patient's symptoms.

The patient received Inj. Ceftriaxone 1 g twice a day for five days post-sclerotherapy. No ureteral stent was placed after the procedure. The postoperative period was uneventful, with no significant adverse effects.



[Table/Fig-1]: Left Retrograde Pyelogram (RGP) showing pyelolymphatic fistula (Arrow).

The patient has been on monthly follow-ups for a year with no recurrence of symptoms or significant complications.

DISCUSSION

Chyluria, or the presence of chyle in the urine, is often caused by abnormal retrograde pyelo-lymphatic channels that link the intestinal lymphatic system with the urinary system. Chyluria is a rare clinical entity, described by ancient scholars like Hippocrates and Galen around 400 BC [1]. The great Indian sage Charak referred to it as 'suklameha' in 300 BC [2]. It is endemic in Southeast Asia, India, China, and parts of Africa. Chyluria can be classified as tropical or non-tropical, and parasitic or non-parasitic. In tropical regions, over 95% of cases are caused by parasitic infestations, primarily by Wuchereria bancrofti. Mosquitoes from the genera Anopheles, Culex, and Aedes transmit this parasite and thrive in waterlogged areas [3].

Non-parasitic causes are rare and are mainly described in case reports, including pregnancy, congenital obstruction, lymphangioma, trauma, radiation, malignancy, and postoperative conditions [4-7]. Mahmood K et al., reported a series of cases of chyluria during pregnancy over a 10-year period, highlighting the hormonal and haemodynamic influences that contribute to lymphatic obstruction during gestation [4]. Dalela D has comprehensively discussed the diagnostic dilemmas and range of aetiologies associated with chyluria, emphasising the importance of distinguishing between parasitic and non-parasitic causes [5]. Krstić TL et al., described a case of congenital lymphatic malformation in a young girl with recurrent chyluria, while Sivashankar M and Nandasena ACN demonstrated successful endoscopic sclerotherapy in an adult male with a lympho-urinary fistula [6,7].

The condition can be classified into mild, moderate, and severe cases. In mild cases, there are intermittent episodes of chyluria. In moderate cases, there is persistent chyluria, with or without colicky pain due to clots, whereas severe cases include systemic symptoms and haematochyluria [8]. Chyluria is essentially a clinical diagnosis, with laboratory tests confirming it. The patient's presentation of chyluria with chyle clots and weight loss indicated a more severe disease. Chyluria is confirmed by a urinary sedimentation test, where a sample is left to settle in a test tube. It separates into three layers: a fatty upper layer, a fibrinous middle layer, and a sediment lower layer [9].

In this case, the detection of chyluria was performed physically by observing the urine sample as well as examining sediment in the test tube and under microscopy. There was no evidence of filaria detected in the blood smear, and cystoscopy showed left-sided involvement. Intravenous urography, retrograde pyelography, and lymphangiography typically demonstrate lymphatic-urinary fistulas, which could be therapeutic in some cases due to contrast-induced chemical pyelitis [10]. Lymphangiography is considered the gold standard and can demonstrate the site, calibre, and number of fistulas; however, it is invasive, time-consuming, technically demanding, and is rarely used nowadays [11]. Chyluria has a uniquely unpredictable course of disease remission and recurrence.

Renal pelvic instillation of sclerosing agents is a treatment modality that induces an inflammatory response in the lymphatic vessels, leading to subsequent fibrosis. Silver nitrate and povidone-iodine are the most commonly used agents for sclerotherapy, with alternatives including 10-25% sodium iodide, 50% dextrose, and 76% urografin [11]. Singh KJ et al., highlighted the efficacy of sclerotherapy with various agents, including Iohexol, for treating chyluria. They found that sclerotherapy is a safe and effective alternative to more invasive surgeries, with high success rates regarding symptom resolution and low recurrence rates [12]. Sunder S et al., conducted a study on a series of cases of milky urine and emphasised the importance of timely diagnosis and the role of sclerotherapy in managing this condition. They reported successful outcomes with a variety of sclerosants, including silver nitrate and Iohexol, and stated that the choice of agent should be tailored according to the patient [9].

In patients with failed medical management, surgery is a mainstay of treatment. The procedures performed include chylo-lymphatic disconnection, retroperitoneal lymph-venous anastomosis, autotransplantation, or nephrectomy [3]. In this case, using Iohexol as a contrast agent served a dual function, acting both as a diagnostic tool for the RGP and as a sclerosant, leading to complete relief of symptoms.

CONCLUSION(S)

This case report sheds light on the use of Iohexol as a sclerosant for chyluria. It can serve both diagnostic and therapeutic purposes. It is safe, regularly used for retrograde pyelography, and effective in the treatment of chyluria. Follow-up studies with a larger number of patients need to be conducted to establish protocols and definitive management plans for sclerotherapy in the management of chyluria.

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