International Medical Case Reports Journal

Dovepress

open access to scientific and medical research

Open Access Full Text Article

CASE SERIES

# Rare Cases of Filarial Chyluria in Children

Shetanshu Srivastava[1], Vandana Tiwari[2], Manodeep Sen[3]

[1]Department of Pediatrics, Dr. Ram Manohar Lohia Institute of Medical Sciences Lucknow, Lucknow, UP, India; [2]Department of Biochemistry, Dr. Ram Manohar Lohia Institute of Medical Sciences Lucknow, Lucknow, UP, India; [3]Department of Microbiology, Dr. Ram Manohar Lohia Institute of Medical Sciences Lucknow, Lucknow, UP, India

Correspondence: Shetanshu Srivastava, Department of Pediatrics, Dr RML Institute of Medical Sciences, Vibhuti Khand Gomtinagar Lucknow, 226010, Lucknow, UP, India, Email drsitanshu@yahoo.co.in

**Background:** Lymphatic filariasis leading to the passage of white urine or chyle is a rare manifestation in children. Filarial parasite infiltration leading to abnormal lymphatic–urinary communication occurs with prolonged infection. The incubation period ranges from 5 to 20 yrs., thus relatively infrequent in the pediatric age group. Index of suspicion should be high when a child presents with the passage of white urine because the subclinical manifestation of filarial infection is difficult to recognize. Moreover, more pathognomonic clinical manifestations such as lymphoedema or hydrocoele are present in adulthood. It should also be differentiated from non-parasitic causes like nephrotic syndrome, urates and phosphates in urine, and congenital lymphatic-urinary communication.

**Case Presentation:** We report two pediatric cases with the intermittent passage of milky white urine since one year. Institutional ethical committee approved the study. In both patients, urine triglycerides were high, and the presence of positive filarial antigen test confirmed the diagnosis. Medical management showed remission of symptoms. Our cases highlight the rare presentation of LF in children and the use of point of care diagnostic tests, management, and outcome in them.

**Conclusion:** LF is a rare condition in children, and the index of suspicion should be high for early management.

**Keywords:** chyluria, filaria

## Introduction

Lymphatic filariasis (LF) is a mosquito-borne tropical disease that causes significant disability. The World Health Organization (WHO) estimated that LF is found in 81 tropical and subtropical countries with 120 million infected cases and one billion people at risk; 947 million people are threatened, whereas this infection disfigures 40 million people. [1] About 90% of the estimated 120 million cases of LF in the world are caused by the filarial parasite Wuchereria bancrofti.[2] In children, microfilariae nest in the lymphatic system post-infection remains subclinical for years. It gives rise only to non-specific presentations of adenitis/adenopathy hence difficult to recognize. However, after puberty, adult disease syndromes like lymphoedema and hydrocoele manifest.[3] Recognizing LF by atypical presentation like chyluria in children and timely diagnostic tests have therapeutic and preventive implications.

## Novelty

Our cases highlight rare clinical presentation of LF in children along with the use of newer point of care diagnostic tests and management strategies.

## Case Presentation

We report two cases who presented in the pediatric outpatient Department of a tertiary care hospital with the intermittent passage of milky white urine after obtaining written informed consent for publication of their details from the patient kin. Institutional ethical committee of Dr Ram Manohar Lohia Institute of Medical Sciences approved the study and granted approval to publish the case details.

Case 1: an 8-year girl with a history of undocumented fever for one year with the passage of white urine. There was no accompanying abdominal pain, dysuria, hematuria, decreased urine output, edema, or burning micturition. There was no

Received: 16 September 2021
Accepted: 9 March 2022
Published: 30 March 2022

aggravating or relieving factor, no history of abdominal trauma. On examination, mild pallor was present, the physical examination was normal.

## Investigations

Blood serum chemistry showed normal complete blood counts (hemoglobin 10.5g/dL, white blood cell 7x109/L, platelet count 250 x10^9/L).Eosinophil count normal. The peripheral blood film showed microcytic hypochromic anemia with no parasites. Renal function tests and serum electrolytes were normal. Serum albumin level was 3.2 g/dL (normal range: 3.5–5.5 g/dL), total cholesterol level was 180mg/dL (normal range: 120–220 mg/dL), and serum triglycerides were 128 mg/dL (normal range: 80–150 mg/dL). Urine was milky white in color (Figure 1) and albumin was (2+), urine triglycerides was 140 mg/dl, which was high. 24-hour urinary protein was 1.5 gm/day. The absence of nephrotic range proteinuria, edema, hypoproteinemia, and hypercholesteremia ruled out nephrotic syndrome. Urine did not clear on heating and on adding 10% acetic acid suggesting the absence of urates and phosphates, respectively. Timing of symptoms and ultrasonography of the abdomen ruled out any congenital causes. Furthermore, since urine triglycerides were high, we did a blood examination for filarial antigen (CFA), which was positive.

Case 2: 10 years old girl presented with poor appetite and not gaining weight. She revealed intermittent passage of white urine since one year. She was afebrile with a normal systemic examination. Her weight for age was 20 kg (less than 3 SD for age), BMI was 12.5, which was less than 3rd centile for age.[4] Investigations showed normal blood counts. Peripheral smear was also normal. Renal function tests, serum albumin and Serum cholesterol level, serum electrolytes were normal. Urine was milky white (Figure 2). Urine protein was negative, and urine triglycerides was high at 135mg/dl, which raised the suspicion for filaria. After ruling out nephrotic syndrome, we did a filarial antigen (CFA) test, which was positive. It is a very sensitive test for diagnosing Wuchereria bancrofti infections.

## Treatment

The etiology of chyluria was filaria in both cases, which showed remission after treatment with diethylcarbamazine (DEC) (6 mg/kg/day) for 21 days. The urine had completely cleared.

Dietary modifications of low fat and high protein diet (2–4 gm/kg) were done along with medium-chain triglycerides (MCT) supplementation.Fat-soluble vitamins ADEK were given inappropriate doses.

## Advice to Parents

Since it is a mosquito-borne disease, proper measures to prevent mosquito bites are advised. Urine had completely cleared after two weeks. Antigen tests done at six weeks were negative for microfilariae. Both children were afebrile and also showed improvement in their appetite. Follow-up was done for six months. There were no recurrences.

**Figure 1** White urine/chyluria (Case 1).

**Figure 2** Chyluria (Case 2).

## Discussion

In children, intermittent chyluria is often missed. They are investigated and treated for common conditions like nephrotic syndrome characterized by the passage of white urine due to loss of significant amount of proteins, which is more than 1gm/m2/day.[5] Index of suspicion should be high for filaria, and timely diagnosis is important for early treatment. There are only a few case reports in children of lymphatic filariasis.[5–8] Differentiating from nephrotic syndrome is necessary to avoid unnecessary renal biopsies[9,10] and treatment.There are reports[11] of adult patients having chyluria who were misdiagnosed and aggressively treated with immunosuppressive drugs. Other causes of white urine are the presence of urates and phosphates, congenital abnormal lymphatic-urinary communication, and post abdominal trauma.[12,13] White urine having urates clears on heating, and phosphaturia clears on adding acetic acid.[8] Our patients tested negative for both. Chyluria can be diagnosed by the presence of triglycerides in urine, as in our patient. Other urine tests are mixing a milky urine sample with equal amounts of ether, which clears it, and oral ingestion of fat labeled with Sudan III, which causes orange, pink discoloration of urine.[14] We did not do these tests in our patients.

Chyluria occurs due to intestinal lymph drainage following dilatation and rupture into the urinary tract due to obstruction.[15,16] Congenital abnormalities of the lymphatic vessels, malignancy, lymph urinary fistulas secondary to renal injuries are causes of non-parasitic chyluria.[17–19] Most cases of parasitic chyluria are by Wuchereria bancrofti filariasis.[1]

## Transmission

Adult worms nest in the lymphatic vessels for 6–8 years, disrupting its normal function. Subsequently, they produce millions of immature larvae or microfilariae, which circulate in the blood. Mosquitoes are infected with microfilariae while taking a blood meal of an infected host which subsequently spreads to others. Mature parasite larvae are deposited on the skin of humans. They then migrate to the lymphatic vessels, thus continuing the transmission cycle.[20] Chyluria is rare in children, with no clear-cut diagnostic approach and management strategies.

## Diagnosis of Filaria

Childhood LF is incompletely documented due to the lack of specific clinical features and limitations of previously available diagnostic methods. Most published surveys based their diagnosis of infection on smear examination. Demonstration of microfilaria in blood is a specific test but lacks sensitivity, especially in children where adult worms are present but no microfilariae.[21] The microfilariae can be detected directly through blood smear examination or DEC-provocative test.

However, in our patients, the smear was negative. Alere Filariasis Test Strip (FTS) – is a qualitative, point-of-care diagnostic tool that detects Wuchereria bancrofti circulating filarial antigen (CFA) in human blood, serum, or plasma.[21–23] Usage of the point-of-care rapid diagnostic test is essential for early detection.

Dietary therapy is of paRamount importance in children with chyle leaks as it aims to improve nutritional status and maintain hydration and electrolyte balance also decrease chyle production.[24]

As per available literature,[24,25] dietary modifications like fat-free/very low-fat diet (a diet containing <0.5 g fat per serving), increased intake of MCTs like coconut oil and a high-protein diet is beneficial. Medium-chain triglycerides (MCT) are recommended to treat chyle leaks. Consumption of cereals, fruits, and vegetables would further improve the nutrition status of children with chyluria. Nutritional status must be monitored; fat-soluble vitamin and essential fatty acid supplementation is of paramount importance.

## Conclusion

Both our patients had chyluria due to lymphatic filariasis, which is rare in children. A high index of suspicion is required regarding its clinical manifestations. Circulating filarial antigen-(CFA) is an important point of care test. Proper Medical management is required for complete remission and to prevent recurrences, along with dietary management.

## Disclosure

The authors report no conflicts of interest in this work.

## References


1. Global programme to eliminate lymphatic filariasis: progress report, 2020 WHO. Lymphatic Filariasis. Geneva: WHO; 2020. Available from: https://www.who.int/news-room/fact-sheets/detail/lymphatic-filariasis. Accessed March 17, 2022.
2. World Health Organization. Global programme to eliminate lymphatic filariasis: progress report for 2018. *Wkly Epidemiol Rec*. WER No 41. 2019;41:457–472.
3. Witt C, Ottesen EA. Lymphatic filariasis: an infection of childhood. *Trop Med Int Health*. 2001;6(8):582–606. doi:10.1046/j.1365-3156.2001.00765.x.
4. Khadilkar V, Yadav S, Agrawal KK. Revised IAP Growth Charts for Height, Weight and Body Mass Index for 5- to 18-year-old. *Indian Children Indian Pediatr*. 2015;52:47–55. doi:10.1007/s13312-015-0566-5
5. Garg G, et al. Chyluria in young child: a rare presentation. *BMJ Case Rep*. 2018;16:bcr–2017. doi:10.1136/bcr-2017-223516
6. Das PK, Sirvidya A, Vanamail P, et al. Wuchereria bancrofti microfilaraemia in children in relation to parental infection status. *Trans R Soc Trop Med Hyg*. 1997;91(6):677–679. doi:10.1016/S0035-9203(97)90521-6
7. Stalens JP, Falk M, Howmann-Giles R, et al. "Milky" urine—a child with chyluria. *Eur J Pediatr*. 1992;151(1):61–62. doi:10.1007/BF02073895
8. Kohli V, Gulati S, Kumar L. Filarial chyluria. *Indian Pediatr*. 1994;31:451–454.
9. Gulati S, Gupta N, Singh NP, et al. Chyluria with proteinuria or filarial nephropathy? An enigma. *Parasitol Int*. 2007;56(3):251–254. doi:10.1016/j.parint.2007.03.002
10. Cheng J-T, Mohan S, Nasr SH, et al. Chyluria presenting as milky urine and nephrotic-range proteinuria. *Kidney Int*. 2006;70(8):1518–1522. doi:10.1038/sj.ki.5001703
11. Kaul A, Bhadhuria D, Bhat S, et al. Chyluria: a mimicker of nephrotic syndrome. *Ann Saudi Med*. 2012;32(6):593–595. doi:10.5144/0256-4947.2012.593.
12. Diamond E, Schapira HE. Chyluria—a review of the literature. *Urology*. 1985;26:427–431. doi:10.1016/0090-4295(85)90147-5
13. Sharma S, Hemal AK. Chyluria-An overview. *Int J Nephrol Urol*. 2009;1:14–26.
14. Liu DY, Wang HF, Xia WM, et al. right-crossed, fused renal ectopia L-shaped kidney type with urinary chyluria. *Urol Int*. 2015;95:243–245. doi:10.1159/000382132
15. Singh K, Kumar M, Jhanwar A, et al. Crossed fused renal ectopia with chyluria: a rare presentation. *BMJ Case Rep*. 2016;2016:bcr2015213050. doi:10.1136/bcr-2015-213050
16. Kekre N, Arun N, Date A. retroperitoneal cystic lymphangioma causing intractable chyluria. *Br J Urol*. 1998;81:327–328. doi:10.1046/j.1464-410X.1998.00337.x
17. Kano K, Arisaka O. Chyluria due to retroperitoneal lymphangioma producing nephrotic syndrome. *J Pediatr*. 2003;143:685. doi:10.1067/S0022-3476(03)00288-9
18. Eisner BH, Tanrikut C, Dahl DM. Chyluria secondary to lymphorenal fistula. *Kidney Int*. 2009;76:126. doi:10.1038/ki.2009.64
19. Van Olden RW, van Dam FE, Kho S, et al. chyluria caused by congenital retroperitoneal lymphangiectasis. *Ned Tijdschr Geneeskd*. 1989;133:1701–1703.
20. Lymphatic Filariasis: Biology - Life Cycle of Wuchereria bancrofti. CDC; 2018. Available from: https://www.cdc.gov/parasites/lymphaticfilariasis/biology_w_bancrofti.html. AccessedMarch 25, 2022.
21. Weil GJ, Lammie PJ, Weiss N. The ICT ®lariasis test: a rapid-format antigen test for diagnosis of Bancroftian ®lari- asis. *Parasitology Today*. 1997;13:401–404. doi:10.1016/S0169-4758(97)01130-7
22. Chesnais CB, Vlaminck J, Kunyu-Shako B, et al. Measurement of Circulating Filarial Antigen Levels in Human Blood with a Point-of-Care Test Strip and a Portable Spectrodensitometer. *Am J Trop Med Hyg*. 2016;94(6):1324–1329. doi:10.4269/ajtmh.15-0916.
23. Gass K, de Rochars MVB, Boakye D, et al. A Multicentre evaluation of diagnostic tools to define endpoints for programs to eliminate bancroftian filariasis. *PloS Negl Trop Dis*. 2012;6(1):e1479. doi:10.1371/journal.pntd.0001479
24. Mccray S, Parrish CR. When chyle leaks: nutrition management options. *Pract Gastroenterol*. 2004;2:60–77.
25. Hashim SA, Roholt HB, Babayan VK, et al. treatment of chyluria and chylothorax with medium-chain triglyceride. *N Engl J Med*. 1964;270(15):756–761. doi:10.1056/NEJM196404092701502