CASE REPORT

Chyluria in young child: a rare presentation

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SUMMARY

Chyluria, a chronic manifestation of lymphatic filariasis, is rare in children. Clinicians must have a high index of suspicion to diagnose this condition in children as it mimics nephrotic syndrome. We present an unusual case in which a 7-year-old boy hailing from a filarial endemic region presented with a passage of milky urine, which on evaluation was diagnosed as parasitic chyluria. The child showed remission after medical management that persisted until 1 year of follow-up.

BACKGROUND

Chyluria or passage of 'milky urine' occurs due to the presence of chyle in urine as a result of abnormal lymphatic-urinary communication.¹ Chyluria is one of the chronic manifestations of lymphatic filariasis that usually has a long incubation period ranging from 5 to 20 years.² It is, therefore, rare in children. We report an unusual case of a 7-year-old child who presented with chyluria; the aetiology turned out to be filarial and it showed complete remission after medical management.

CASE PRESENTATION

A 7-year-old boy who was native of a north-east part of India presented with an intermittent passage of milky urine and bilateral pedal oedema for the last 3 months. The child gave a history of few episodes of haematuria with renal colics. The child was fairly built with body mass index (BMI) of 16.5 kg/m² (above 50th percentile WHO reference BMI for age).³ The vital signs were stable (blood pressure 98/56 mm Hg, pulse rate 96/min). There was no pallor or inguinal lymphadenopathy; chest and abdominal examinations were normal. Both legs showed prominent pitting oedema. There were no signs of essential fatty acid/fat-soluble vitamin deficiency (skin changes, hair depigmentation, and so on).

INVESTIGATIONS

Blood serum chemistries revealed normal complete blood counts (haemoglobin 11 g/dL, white blood cell 9×10^9 /L, platelet count 250×10^9 /L), normal renal function tests (blood urea 25 mg/dL, serum creatinine 0.9 mg/dL) and serum electrolytes (sodium 136 mEq/L, potassium 4.4 mEq/L). Serum albumin level was 2.2 g/dL (normal range: 3.5–5.5 g/dL), total cholesterol level was 266 mg/dL (normal range: 120–220 mg/dL), and serum triglycerides level was 229 mg/dL (normal range: 80–150 mg/dL). Urinalysis showed the presence of protein (4+) using a dipstick test, 4–6 red blood cells/high power

field, massive proteinuria (2 g/24 hours) and presence of fat globules confirmed by Sudan stain III. Microfilariae antigen test-immunochromatographic test (ICT card test) was positive. Ultrasonography of abdomen was unremarkable. The diagnosis of parasitic chyluria was made.

DIFFERENTIAL DIAGNOSIS

1. chyluria
2. nephrotic syndrome
3. urinary tract infection
4. phosphaturia.

TREATMENT

The child was given drug treatment for filariasis: a course of diethylcarbamazine (DEC; 6 mg/kg/day) for 21 days along with dietary modifications. In consultation with a dietitian, the child was given an enteral fat-free (fat contributing less than 15% total calorie intake) and high-protein (1 g/kg/day) diet. The child also received oral multivitamin supplements containing fat-soluble vitamins. After discharge from the hospital, the parents of the child were advised to prevent mosquito bites. They were also given the following broad instructions pertaining to diet: the amount of fat in the diet should be as low as possible (use of medium-chain triglycerides (MCT) containing foods mainly); and ensure adequate intake of cereals, pulses, fruits and vegetables. The fat proportion in the diet was gradually increased (long-chain triglycerides containing foods) after 3 months and the child resumed normal diet in the next 6 months.

OUTCOME AND FOLLOW-UP

At the end of 10 days, the urine had completely cleared. At the end of 4 weeks, the blood tests were negative for microfilariae (ICT card test). The child showed normal growth kinetics (BMI for age: 18.5 kg/m²; above 85th percentile WHO reference range) and also did not report any recurrence of chyluria at the end of 12-month follow-up.

DISCUSSION

Lymphatic filariasis continues to be a major health problem in many tropical countries. Some common chronic manifestations of lymphatic filariasis include vaginal hydrocoele, lymphoedema and chyluria.⁴ In children, chyluria is usually non-filarial either due to congenital abnormalities of lymphatics, lympho-urinary fistulas secondary to renal injuries and lymphatic obstruction.^{4–6} In the present case, the cause of chyluria turned out to be filarial

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Table 1 Illustration of available case reports and comparison with present case

Author	Age (years)	Cause	Treatment received	Follow-up (months)
Stalens <i>et al</i> ⁹	10	Congenital pyelovesical communication	Conservative	–
Kohli <i>et al</i> ¹⁰	10	Parasitic	Conservative	1
Jung <i>et al</i> ¹¹	10	–	Conservative	10
McNeil <i>et al</i> ¹²	5	Malignancy (metanephric adenoma)	Surgery (nephrectomy)	12
Ganesh <i>et al</i> ¹³	5	–	Surgery (laparoscopic lymphorenal disconnection)	6
Present case	7	Parasitic	Conservative	12

as the ICT test was positive for microfilariae. In children, chyluria is often confused with the clinically more common condition of nephrotic syndrome as both conditions are characterised by anasarca, malnutrition with the milky white appearance of urine secondary to loss of significant amount of proteins (>3 g loss in 24 hours).⁷ Differentiating between the two is necessary as the management of these conditions is different and it can avoid unnecessary renal biopsies and immunosuppressive drugs. A study reported that 12 adult patients (average age: 34.5 years) were misdiagnosed with nephrotic syndrome and were initially treated with immunosuppressive drugs with no benefit and were eventually found to have chyluria.⁷ There are various clues that can help decide whether the child is having chyluria or nephrotic syndrome. For example, haematuria due to rupture of blood vessels near the abnormal fistulous connections and renal colic due to clots can occur in chyluria but is not seen in classical nephrotic syndrome.² Children can rarely acquire chyluria, especially in the endemic areas. Das *et al* reported that familial clustering of microfilariae can occur in children due to repeated exposure.⁸ In this subset of patients, repeated attacks of lymphatic filariasis can lead to irreversible abnormalities in renal lymphatics. The dilatation of lymph vessels and stasis and superimposed acute filarial attacks may lead to the development of chronic manifestations like hydrocoele, lymphoedema or chyluria even in children. After an extensive and thorough search of the medical literature, we could identify only five case reports of chyluria in children (table 1).^{9–13} Kohli *et al* reported a case of microfilaraemic chyluria in a 10-year-old male child from Chandigarh, India.¹⁰ The child was managed with conservative management including DEC (6 mg/kg/day) for 21 days. The authors reported that the child experienced remission and his peripheral blood films became negative for microfilaria after 1 month of starting therapy. However, there was no mention about the further recurrences, if any, in the child.¹⁰ Ganesh *et al* described a case of unilateral chyluria in a 5-year-old child hailing from South India who presented with resistant chyluria not responding to medical therapy and was managed with laparoscopic lymphorenal disconnection.¹³ The authors described that there was no recurrence at 6-month follow-up.¹³ As chyluria in children is very rare, there is no consensus on dietary modifications in chyluria. The goal of dietary therapy in patients with chyle leak is to decrease chyle production, maintain fluid and electrolyte balance along with maintenance/improvement of nutritional status.¹⁴ Using the experience of studies done on dietary modifications in chyle leak syndromes like chylothorax and chylous ascites, we formulated dietary instructions for our patient.^{14 15} A reasonable regimen for this group of patients would be to include a fat-free (diet containing <0.5 g fat per serving) and a high-protein diet.¹⁴ Foods containing MCTs like coconut oil may be used, as MCTs have been

found to get directly absorbed through intestinal mucosa in the portal circulation.¹⁵ Various other dietary measures such as using skimmed milk, avoiding deep-fried foods and ensuring intake of cereals, fruits and vegetables have been also proposed to decrease chyle production and optimise nutrition of patients.¹⁶ For children presenting with heavy chyle leaks, commercial preparations containing MCT, like MCT oil in doses of 60–70 g/day (4–5 table-spoons; ~500–600 calories), along with providing 2%–4% of daily calorie intake in the form of essential fatty acids like safflower, sunflower or corn oil are recommended to prevent essential fatty acid/fat-soluble vitamin deficiency.¹⁴ The child followed the dietary modifications for 3 months after starting treatment.

Although there is the role of dietary restriction/modifications in the initial management of chyluria, its role as long-term management is ambiguous. There are no studies where the impact of the dietary restriction on recurrence on chyluria has been studied. In endemic zones where the disease dynamics is complicated, we are not too rigid on diet modifications as there are so many unknown variables in chyluria (availability of resources, poverty, difficult to avoid further exposure to mosquito bite, difficulty in adherence to diet, and so on). We therefore give broad instructions only. The usual instructions given are: the amount of fat in the diet should be as low as possible; and ensure adequate intake of cereals (roti, rice, broken wheat and bread), pulses, fruits and vegetables. Here, we report a rare unique case of chyluria in a child who hailed from an endemic zone and responded to medical management.

Learning points

- ▶ Chronic manifestations of lymphatic filariasis like chyluria may rarely develop in children especially in the endemic areas.
- ▶ A high index of suspicion is needed to suspect this condition in children hailing from endemic zones and differentiate this from the more common nephrotic syndrome in order to avoid unnecessary treatments.
- ▶ The prognosis of parasitic chyluria seems to be good for children with few recurrences and complete remission with medical management.

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