# A case of a child with non-parasitic chyluria

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Chyluria is the passage of milky urine due to the leakage of lymph into the urinary tract. Chyluria occurs predominantly in adults and is rare in children. We present an unusual case in which a child with proteinuria, hematuria and milky urine was subsequently diagnosed with non-parasitic chyluria. Retrograde cystogram confirmed a lymphatico-calyceal communication. This case showed spontaneous remission. The etiology of this case was not exactly known; however, the prognosis of non-parasitic chyluria (or idiopathic chyluria) is usually very good and the treatment is mostly conservative. **(Korean J Pediatr 2006;49:326-328)** 

Key Words: Child, Conservative treatment, Non-parasitic chyluria

#### Introduction

Chyluria occurs as a result of communicaton between the lymphatics and the urinary system<sup>1)</sup>. Chyle is composed mainly of albumin, chylomicron, and fibrin in various portions. Thus, intermittent mild proteinuria and hematuria may be found at routine urinalysis of chylous urine<sup>2)</sup>. The conventional diagnostic approach involves confirmation of chyle in the urine and the demonstration of lymphaticourinary fistulae by radiological study. A low-fat, high-fiber diet is the initial treatment. Aggressive treatment should be suggested only when general health conditions are seriously compromised. We report the rare case of a child with non-parasitic chyluria.

### Case Report

A 10-year-old male was admitted to the hospital because of milky urine with occasional red gelatinous clots causing urinary retention (Fig. 1). The symptoms abruptly developed before admission. The patient has lived in Seoul and had not traveled to areas known for endemic filariasis. On examination, he was normotensive, with no lympha-

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Tel:02)950-1074 Fax:02)951-1246, 938-4109 E-mail:koojw9@sanggyepaik.ac.kr denopathy, abdominal masses or edema. Urinalysis showed milky color, proteinuria (4 positive), hematuria (4 positive) and leukocyturia (many WBC/HPF microscopically). Urine culture was negative. A 24-hour urine sample contained protein (7,600 mg/day) and triglycerides (7,740 mg/day). Urine electrophoresis revealed severe albuminuria and nonspecific proteinuria. Urine cytospin analysis revealed lymphocyte dominant (88%) fluid and suggested that urine



Fig. 1. At admission, he was passing milky appearing, viscous, pinkish, and turbid urine.



**Fig. 2.** Retrograde pyelogram demonstrating lymphatico-urinary communication on the right kidney. Note the extravasated lymphangiographic contrast material in the right renal calices (arrows).

was mixed with lymphatic fluid. The complete blood count was normal (hemoglobin 13 g/dL, hematocrit 38.4%, WBC 6,540/mm<sup>3</sup>, platelet 283,000/mm<sup>3</sup>) with no evidence of microfilaria or eosinophilia on peripheral smear. We repeated the examinations of thick blood smears taken at midnight, and no microfilaria or acid-fast bacilli was detected in the thick blood films. Biochemical serum analysis showed the following results:blood urea nitrogen 12 mg/dL, creatinine 0.6 mg/dL, sodium 140 mEq/L, potassium 4 mEq/L, protein 6.7 g/dL, albumin 4.2 g/dL. Here the patient had neither hypoproteinemia nor anemia.

Cystoscopy showed chylous efflux from the right ureteral orifice. Retrograde pyelogram showed pyelolymphatic backflow into the renal lymphatic sinuses (Fig. 2). Lymphoscintigraphy was performed three weeks later and was normal. Renal ultrasonography confirmed kidney with normal anatomy. Chest X-ray, renal ultrasonography and abdomino-pelvic CT scan did not demonstrate a cause for the chyluria and other diseases were excluded.

The patient was treated with conservative management (a low-fat, high-fiber diet, abdominal binder) and seven weeks later, the chyluria disappeared. On the 10-month follow-up, the patient remained symptom -free without chyluria.

#### Discussion

In tropical areas, the most common cause of chyluria is filariasis. However, in nonendemic areas, chyluria is more commonly caused by congenital abnormalities of the lymphatic system as well as by inflammatory, pregnancyrelated, traumatic or neoplastic retroperitoneal lymphatic obstruction<sup>3</sup>. In the early part of the century, filariasis was seen in the coastal region in Korea. However, for the past 20 years there has been no evidence to suggest that the disease is endemic in Korea<sup>4</sup>. The exact cause of chyluria in this case was not determined, as the filarial blood smear was negative and no other cause was found.

The finding of lymphocytes and the presence of fat globules in the urine formed the basic diagnostic criteria. The absence of such findings in a single random sample of urine, however, does not rule out the disease in patients with a history of "milky" urine. Further examination for fat globules in urine should be made, especially after a fatty meal. Once the diagnosis is established, the approach lies in assessing the severity of the disease and the site of lymphatico-calyceal fistulisation. The severity of the disease could be guided by the symptomatology such as persistent symptoms, history of clot colic, urinary retention and significant loss of body weight.

Lymphoscintigraphy is a noninvasive, safe and simple technique for investigating the lymphatic system. It is advocated as the method of choice for screening of patients with lymphatic urinary communication<sup>5</sup>. However, its diagnostic accuracy is still inferior to conventional lymphangiography<sup>6</sup>. Lymphoscintigraphy failed to demonstrate the site of communication between the lymphatic vessels and the urinary tract in our case. So we confirmed lymphatico-calyceal communication by retrograde pyelogram.

In this case, the chyluria spontaneously disappeared and the urine became clear and yellowish after retrograde pyelogram. But the chyluria recurred two days later.

The patient was treated with conservative management, i.e. a low-fat, high-fiber diet and an abdominal binder. Seven weeks later after treatment, the chyluria had disappeared. This case is interesting because the passage of milky urine stopped suddenly after the patient wore the abdominal binder for three weeks. As spontaneous remission of chyluria has been reported<sup>77</sup>, we cannot be certain whether in this case the abdominal binder was responsible

for resolution of the chyluria.

As most cases of non-parasitic chyluria showed spontaneous remission, surgical treatment should be restricted to patients with severe symptoms, i.e. weight loss, recurrent clot colic, clot retention, severe anemia, hypoproteinemia or chyluria not responding to conservative treatment and sclerotherapy<sup>8)</sup>.

#### 한 글 요 약

## 비기생충증 원인에 의한 유미뇨 1례

인제대학교 의과대학 상계백병원 소아과, 서울대학교 의과대학 소아과학교실\*

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유미뇨는 비뇨기계의 림프계와 정맥계가 누관으로 연결되어 유미(chyle)가 소변으로 배설되는 것을 말한다. 평상시 건강히 지내던 10살된 남아가 딸기 우유빛 소변을 주소로 내원하여 시 행한 방광경검사로 우측 요관 입구에서 우유색뇨와 혈액이 분출 되는 소견을 보았고, 역행성 신우조영술로 우측신 상부에서 신 우-임파역류 소견을 확인하였다. 저자 등은 비기생충증 원인의 유미뇨를 가진 환아에서 보존적 치료법만으로도 자연 치유된 유 미뇨 1례를 경험하였기에 이를 보고하고자 한다.

#### References

- Lang EK, Redetzki JE, Brown RL. Lymphangiographic demonstration of lymphaticocaliceal fistulas causing chyluria (filariasis). J Urol 1972;108:321–4.
- Rysznyak I, Foldi M, Szabo G. Lymphatics and Lymph circulation, 2nd ed. New York Pergamon, 1967:785–99.
- 3) Lai KN, Siu D, Chan KW, Lai FM, Yan KW. The clinical significance of proteinuria in patients with nonparasitic chyluria. Am J Kidney Dis 1986;7:381–5.
- 4) Paik YH, Cho YJ, Koo DS, Ree HI, Shim JC. Studies on the current epidemiological situation of brugian filariasis in endemic areas of Korea. Korean Parasitol 1988;26:255–62.
- Pettit J, Sawczuk IS. Use of lymphoscintigraphy in chyluria. Urology 1988;32:367–9.
- Haddad MC, al Shahed MS, Sharif HS, Miola UJ. Case report: Investigation of chyluria. Clinical Radiology 1944;49: 137-9.
- Ohyama C, Saita H, Miyasato N. Spontaneous remission of chyluria. J Urol 1979;121:316–7.
- Hemal AK, Gupta NP. Retroperitoneoscopic lymphatic management of intractable chyluria. J Urol 2002;167:2473–6.