İntermitan Şilüri: Olgu Sunumu

Intermittent Chyluria: Case Report

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ÖZET

Lenfatik sıvının idrara karışması şeklinde tanımlanan şilüri nadir görülen bir durumdur. Konjenital veya kazanılmış nedenlerle ortaya çıkan bir tıkanma sonucu lenfatik sistemde basınç artar, böylece renal lenfatik sistem ile pelvikalisiyel sistem arasında fistül oluşur.

Yirmi dokuz yaşındaki kadın hasta, bulanık ve süt renginde idrar yapma yakınmasıyla başvurdu. Öyküsünden bu yakınmasının 1.5 yıldır olduğu ve belirli aralıklarla ortaya çıktığı öğrenildi. Olgunun sabah idrar analizinde; trigliserid 778 mg/dl, protein 396 mg/dl, öğleden sonraki idrar analizinde ise trigliserid 82 mg/dl, protein 4 mg/dl saptandı. Bu bulgularla şilüri tanısı alan olgumuzda, izlemde şilüri kayboldu. Şilüri endemik bölgeler dışında nadir görüldüğü için, bu olgu da ilginç bulunarak sunulmuştur.

Anahtar sözcükler: şilüri, lenfosintigrafi, lenfanjiyografi, lenfatik sistem

ABSTRACT

Chyluria is defined as the presence of lymph in the urine and is rare except in the areas of the world where filariasis is endemic. An abnormal communication or fistula is formed between the renal lymphatic system and urinary collecting system due to high intralymphatic pressure caused by an obstruction. This obstruction may be congenital or acquired.

A twenty-nine-year-old woman presented with a complaint of milky urine lasted for 18 months intermittently that exacerbates and resolves spontaneously. Chemical analysis of urine sampled in the morning showed triglyceride levels of 778 mg/dl, protein levels of 396 mg/dl. Triglyceride level of the urine sampled in the afternoon was 82 mg/dl, and protein level was 4 mg/dl. This prominent hypertriglyceriduria led to the diagnosis of chyluria. During the follow-up period of the patient, chyluria resolved. Chyluria is not commonly seen in non-endemic areas so this case was found to be interesting and has been reported.

Keywords: chyluria, lymphoscintigraphy, lymphangiography, lymphatic system

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Introduction

Chyluria is defined as leakage of lymphatic fluid into the urine as a consequence of fistula formation between the renal lymphatic system and the pelvicalyceal system (1). The first manifestation of chyluria is milky-white urine (2). Chyluria contains high amounts of lipids, albumin and fibrin. Chyluria may result in hypoalbuminemia, hypolipidemia, anemia, malnutrition and abnormalities of the immune system; thus diagnostic and therapeutic methods are important in the case of chyluria (3). The most common causes of chyluria are parasitic infections in the endemic areas and trauma, tuberculosis, congenital lymphatic malformations and retroperitoneal tumors in the non-endemic areas (2,4).

A subject with chyluria was presented in this case report as it was a rare condition.

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Case Report

A 29-year-old woman presented with a complaint of turbid and milky urine. Her history revealed that she had no previously known disease and the symptoms began about 1.5 years ago. The patient told that

she urinated turbid and milky urine when she waked from sleep and had dysuria during urination at times over this period. Her history also revealed that she received antibiotic treatment several times because pathogens were detected from the urinary cultures performed for her symptoms and in addition her results of cystoscopy were found to be normal.

The patient with the complaint of turbid and milky urine that appeared intermittently (with an average interval of 40 to 45 days) was admitted to the hospital. Her physical examination, complete blood count, blood biochemistry and erythrocyte sedimentation rate were all found to be normal. Her morning urine was turbid and milky white in color (Figure 1). Her urinary examination did not reveal leukocyte, erythrocyte, bacteria or acido-resistant bacteria (ARB) and the urinary cytology was found to be normal. No growth (non-specific, fungal or ARB) occurred in the urinary cultures. Chest radiography, abdominal ultrasound and abdominal magnetic resonance imaging (MRI) were found to be normal.

The results of examination of the turbid and milky white urine were as follows: triglyceride 778 mg/dl, total cholesterol 17 mg/dl, protein 396 mg/dl. Urinary lipoprotein electrophoresis have shown alpha lipoprotein at the rate of 4.6% and prebeta 56.6%, beta 27% and chylomicron 11.8%. Triglyceride was found to be 82 mg/dl, total cholesterol 5 mg/dl and protein 4 mg/dl in the analysis of urinary samples obtained in the afternoon. Fifteen days after her admission, the findings of the patient disappeared completely while her investigations were going on (Figure 2) and her biochemical investigations became normal. The patient who was followed up for an additional time in the clinic and whose findings of turbid and milky white urine did not recur was discharged from the hospital with the instruction to come back for performance of lymphangiography and lymphoscintigraphy in case of recurrence of chyluria.

The patient has been followed up in the outpatient department of our hospital and she has not developed a chyluria attack.

Discussion

Rupture or fistula may develop as a consequence of high intralymphatic pressure in the lymphatic vessels draining the renal lymphatic system. This lympho-urinary communication may develop at the level of the calyx, pelvis, ureter or bladder (2). The chyluria that is defined as leakage of the lymphatic fluid into the urine is a rare condition. Chyluria is a result of congenital or acquired lymphatic obstructions. Accordingly, the lymphatics dilate as a result of an obstruction between the intestinal lymphatics and thoracic lymph channels and these dilated lymphatics lead to lympho-urinary connection by leading to the formation of retrograde fistula with the kidneys and their collecting system (2,5). While the causes of chyluria are varied, the most common cause of it in the endemic areas is parasitic infections (2,4). Other causes of chyluria in non-endemic areas are trauma, abdominal and thoracic surgery, pregnancy, genitourinary and gastrointesti-

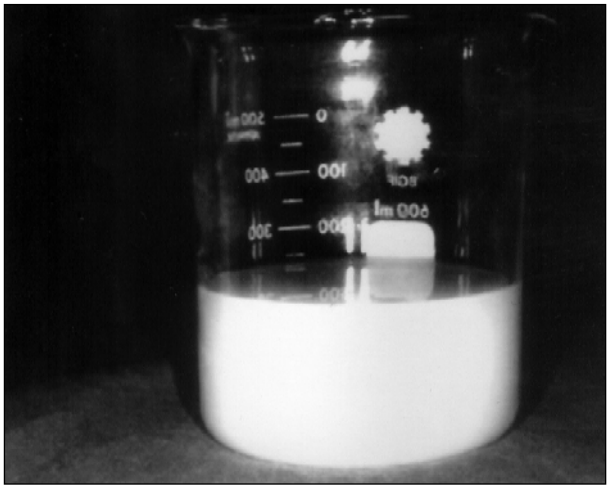

Figure 1. Photograph demonstrates turbid and milky urine.

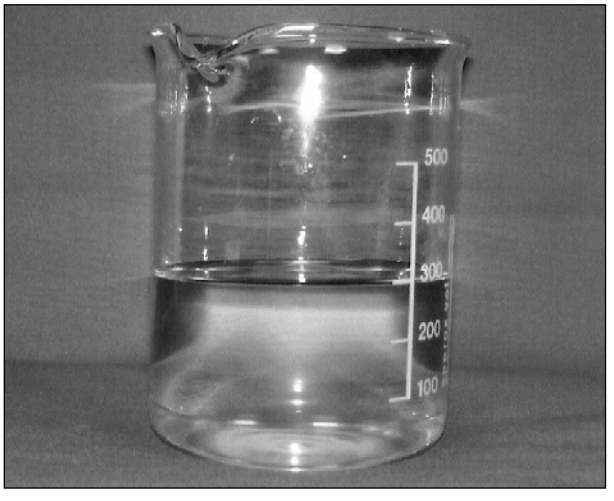

Figure 2. Photograph demonstrates normal urine appearance.

nal tumors, retroperitoneal tumors, or lymphangiectesia, mesenteric adenitis, pelvic lipomatosis, congenital deformities of the thoracic duct, ureter stones, hydrocel, inguinal hernia, chronic inflammatory diseases of the retroperitoneum, tuberculosis, abscess and lymphangiomyomatosis (2,4). In some of the patients, no apparent cause may be detected. We could not find the cause of the chyluria in our patient.

The first manifestation of chyluria is milky-white urine (2). In chyluria, urine may contain high amounts of lipids, albumin, fibrin and varying levels of erythrocytes. Total content of lipids in chyluria may vary depending on the amount of lipids ingested in the diet, the phases of the digestion, type of activity, the position of the patient and whether the patient is sleeping or awake (3,6-8). Chyluria may be continuous or intermittent depending on the changes in the lymphatic obstruction (9). The lymphatic flow is provided mainly by the pressure that is a result of spontaneous and rhythmical contractions of the lymphatic channels (10). Activities and position of the limbs constitute a supporting force for the lymphatic flow (6,10). The lymphatic flow slows down during the resting phase in the night sleep. Protein concentration of the lymphatic fluid rises due to the accumulation during the horizontal position and falls due to accelerated lymphatic flow during upright position (6). Stasis in the lymphatic flow during the sleep may lead the lymphatic fluid to drain into the lymphatic channels of the urinary system and into the collecting system from the intestinal lymphatics abnormally in a retrograde fashion or through the collaterals (6,7,10).

Urinary levels of triglyceride and protein was found to be high in the urinary sample submitted to the laboratory in the morning and to be low in the urinary sample submitted to the laboratory in the afternoon. This suggests that the chyluria in our patient might be related to the increased lymphatic leakage due to the stasis from the slowed lymphatic flow during sleep.

Lymphangiography may be used in investigating the pathology of the lymphatic channels. But this is an invasive diagnostic method. It might lead to local tissue necrosis, fat emboli, hypersensitivity reaction and lymphedema due to the use of contrast agents. Thus, lymphangiography is not widely preferred currently (3,11). Lymphoscintigraphy is a non-invasive, rapid and easy method and does not have any known adverse effects. Favoring results have been reported from the studies assessing the efficiency of this method in detecting the abnormal lymphatic drainage (3,11).

Lymphoscintigraphy did not reveal any pathology in our patient. Because this investigation could not be performed in the period of chyluria attack since the chyluria was intermittent and spontaneously disappeared within 15 days when the patient was in the hospital.

Diet is known to be useful in the course of chyluria known to present with variable clinical course and remission and exacerbations (3,4). Medium-chain fatty acids were known to reduce the formation of intestinal lymph and the lymphatic flow to the thoracic duct (3,4).

In conclusion, chyluria is a rare condition in the non-endemic areas. It may not be possible to find the cause of the chyluria in some patients. Chyluria may result in hypoalbuminemia, hypolipidemia, anemia, malnutrition and abnormalities of the immune system (3); thus diagnostic and therapeutic methods are important in the case of chyluria.

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