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Case report

A rare case of *Mycobacterium fortuitum* infection causing chyluria

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ABSTRACT

We report a case of chyluria caused by *Mycobacterium fortuitum* infection in a sixty-four year old male, who was successfully treated with two weeks of amikacin, trimethoprim-sulfamethoxazole and levofloxacin followed by twenty four weeks of levofloxacin and doxycycline.

Introduction

Chyluria is reflux of intestinal lymph into the urinary colleting system through fistulous communication secondary to obstruction of the retroperitoneal lymphatics, and can be caused by infective or non-infective etiologies. Parasitic infection, in particular filariasis, is the most common infective cause in endemic areas [1,2], while *Mycobacterium tuberculosis* is an uncommon but important infective cause [2]. To the contrary, non-tuberculous *Mycobacterium* (NTM) is a rarely reported cause of chyluria in either immunocompetent or immunocompromised individuals. Here we report a case of chyluria which was found to be caused by *Mycobacterium fortuitum* infection.

Case report

In June 2020, a 64 year old male was referred to the Infectious Diseases Day Ward at the Queen Elizabeth Hospital Hong Kong, for 3-month history of intermittent milky urine and weight loss of 6 kg. He had history of Kallmann syndrome (idiopathic hypogonadotropic hypogonadism), diabetes mellitus, hypertension and hyperlipidemia. There was no associated fever, night sweat, hematuria, loin pain or chest symptoms. He had no history of tuberculosis infection, or surgery / instrumentation to the urinary tract, and had no history of travel or residence in filaria-endemic countries. Other than general muscle wasting, there were no palpable lymph node or abnormal skin lesion on physical examination.

Urine collected was milky in appearance (Fig. 1) and was tested positive for chyle. Bacterial culture was negative while cytological examination showed lymphocyturia. Blood smears were negative for microfilariae. Four early morning urine specimens collected over a three-week period were negative on Ziehl-Neelson stain, but all yielded positive growth of acid-fast bacilli (AFB) after one to two weeks of incubation. MPB64 antigen test (a lateral flow immunochromatographic assay that detects the *Mycobacterium tuberculosis* complex-specific antigen MPB64) was negative. All isolates were subsequently confirmed to be *Mycobacterium fortuitum*, which was susceptible to amikacin, imipenem, ciprofloxacin, trimethoprim-sulfamethoxazole (SXT), doxycycline and linezolid, intermediately resistant to cefoxitin and resistant to clarithromycin. Blood specimen was negative for AFB culture, while chest X-ray did not show any infiltration to suggest active infection. Lymphopenia of 0.4 – 1.1 × 10^3 cells/uL was noted while renal function test and C-reactive protein were normal. Anti-HIV antibody was negative. PET-CT scan of whole body showed the absence of structural abnormality of the urinary tract, abnormal mass, lymphadenopathy or focal FDG uptake.

Once the susceptibility test result was available, treatment was started using intravenous amikacin 10 mg/kg daily, oral SXT 960 mg twice per day and oral levofloxacin 500 mg daily. After 2 weeks of three-drug treatment, regimen was switched to two-drug treatment with levofloxacin 500 mg daily and doxycycline 100 mg twice per day for another 24 weeks. SXT was substituted due to suspected drug-related hyponatremia. Medium chain triglyceride diet was also

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Fig. 1. Milky urine passed by patient.

advised. In view of lymphopenia, CD4/CD8 subsets were checked which were low at 78 and 75 cells/uL respectively. Anti-interferon-gamma antibody was tested negative.

During the 6-month course of antimicrobial treatment till mid-December 2020, chyluria resolved clinically and multiple urine cultures collected after day 12 of treatment were negative for AFB. Four months after treatment completion, there was no recurrence of chyluria clinically, and urine cultures for AFB were repeatedly negative.

Discussion

Mycobacterium fortuitum is ubiquitous in the environment and has been isolated in soil and water [3]. Human infection caused by *Mycobacterium fortuitum* often presented as disseminated infection in immunocompromised hosts, catheter or device-related infections, skin or wound infections, as well as pulmonary infection [4,5]. However, our patient did not present with any of these common scenarios; in particular, the absence of fever, a lack of other infective focus clinically and a negative AFB blood culture did not suggest a diagnosis of disseminated disease. As a result, despite the finding of *Mycobacterium fortuitum* in urine samples, investigations including PET-CT and blood smears were also carried out to rule out the more common parasitic or surgical causes of chyluria. A diagnosis of *Mycobacterium fortuitum* infection was established after a negative workup for the common causes of chyluria, together with the finding of the same isolate in multiple urine samples which meant that contamination was unlikely. However, chyluria, which is resultant from the obstruction of retroperitoneal lymphatics leading to fistulous communication with the urinary collecting system, is a rare clinical manifestation of both tuberculous and non-tuberculous mycobacterial infection. In the literature, there was case report of disseminated tuberculosis presenting with chyluria, which was the result of enlarged lymph node compressing the thoracic duct [2]. In our patient who had chyluria as the sole presenting symptom, the mechanism by which the *Mycobacterium fortuitum* infection causing retroperitoneal lymphatics obstruction was indeterminate from the workup performed. One postulated mechanism would be retroperitoneal lymphatic involvement due to ascending infection from urinary tract. Another possible mechanism would be lymphatic dissemination of infection from an unknown primary site of infection, since the CD4 and CD8 lymphopenia might indicate possible defect in the cell-mediated immunity, hence the predisposition to disseminated mycobacterial infection.

Although *Mycobacterium fortuitum* is more antimicrobial-susceptible than other NTM, the optimal treatment regimen and duration is still unknown [6]. Despite the lack of evidence of disseminated disease both clinically or on whole body imaging, with the finding of the lymphopenia which might suggest possible underlying immunodeficiency, we decided to offer two weeks of initial therapy with a three-drug regimen using amikacin, levofloxacin and SXT, followed by six months of two-drug treatment using levofloxacin and doxycycline to ensure microbiological clearance. Except for mild hyponatremia due to SXT, the antimycobacterial treatment was tolerated and patient was treated successfully with resolution of chyluria and microbiological clearance. In fact, the diagnosis of *Mycobacterium fortuitum* infection was also supported by the fact that symptom resolved completely after starting antimycobacterial treatment. Regarding the CD4 and CD8 lymphopenia, the clinical significance was uncertain at this juncture, since the patient did not had any past episode of opportunistic infection to suggest a full-blown defect in the cell-mediated immunity.

To the best of our knowledge, this is the first reported case of chyluria caused by *Mycobacterium fortuitum* infection. This case illustrated that *Mycobacterium fortuitum* infection is a rare but possible cause of chyluria, and should be considered in any patient in whom the more common parasitic or surgical causes of chyluria have been ruled out.

Consent

Consent could not be obtained from patient as patient was deceased due to other acute illness by the time decision was made to write up the case report.

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Conflict of Interest

The authors report no conflict of interest.

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