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Case Report

Uro-lymphatic fistula associated with urolithiasis: A case report^{☆,☆☆}

Keisuke Tsuboi^{a,*}, Tetsuya Katsumori^a, Daichi Mino^a, Hiroaki Kubota^a, Tatsuya Yoshikawa^b

^aDepartment of Radiology, Saiseikai Shiga Hospital, Ohashi 2-4-1, Ritto, Shiga, 520-3046, Japan

^bDepartment of Radiology, Kyoto Prefectural University of Medicine, 465 Kajii-cho, Kawaramachi-Hirokoji, Kyoto, 602-8566, Japan

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ABSTRACT

Uro-lymphatic fistulas are rare, and involve communication between the renal collecting system and the lymphatic system. The disorder is usually caused by the obstruction of lymphatic vessels due to several diseases, leading to chyluria. Here, we report the case of a patient with a uro-lymphatic fistula, considered to be associated with urolithiasis.

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Introduction

Uro-lymphatic fistulas are rare disorders involving communication between the renal collecting system and the lymphatic system. In most cases, these fistulas are clinically accompanied by chyluria [1]. The disorder is usually caused by parasitic renal or lymphatic infections, including filariasis, echinococcosis, cysticercosis, ascariasis, malaria, and renal tuberculosis [2]. However, the presence of a uro-lymphatic fistula secondary to urolithiasis is rare, and few reports exist in the literature. Here, we report the case of a patient with

a uro-lymphatic fistula considered to be associated with urolithiasis.

Case report

A 58-year-old woman with no history of medical abnormalities presented to our emergency department, complaining of severe pain in the right lower quadrant of the abdomen. Urinary examination showed an elevated level of erythrocytes, suggestive of hematuria; however, no abnormal findings

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* Corresponding author.

E-mail address: tsuboi@koto.kpu-m.ac.jp (K. Tsuboi).

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Figure 1 – A plain computed tomography scan showed a high-density area 5 mm in size (arrow), indicating calculus at the right urinary duct with dilation of the right renal calyx and pelvis.

indicating chyluria were observed. No abnormalities were found on laboratory examinations. Plain computed tomography (CT) showed a 5-mm calculus at the right urinary duct with dilation of the right renal calyx and pelvis (Fig. 1). A physician diagnosed the disorder as right urinary calculosis and hydronephrosis.

The patient's pain persisted despite treatment with analgesics, and she thus returned to the hospital later that same day. Contrast-enhanced CT was performed to exclude several vascular diseases, including dissection or obstruction of major arteries. Although no abnormality of major vessels was found on the arterial and venous phases of contrast-enhanced CT, the delayed phase showed thin vessels around the renal artery/vein that were communicating between the right urinary system and lymph vessels, suggesting a uro-lymphatic fistula localized in the upper pole of the right kidney (Fig. 2A and B).

The patient was conservatively managed with analgesics. Six days after the onset of symptoms, plain CT revealed neither urolithiasis nor hydronephrosis. This finding suggests the spontaneous removal of urinary stones. The patient was then free from clinical symptoms indicating urolithiasis recurrence.

Discussion

In this case, urine directly flowed into lymph vessels through a uro-lymphatic fistula, observed on contrast-enhanced CT. The uro-lymphatic fistula was considered to be associated with obstruction of the urinary system. This is a unique condition, as most uro-lymphatic fistulas are caused by obstruction of lymph vessels; in such cases, lymph fluid flows into the urinary system, leading to chyluria [1]. The patient described here did not have chyluria. This lack of chyluria is considered re-

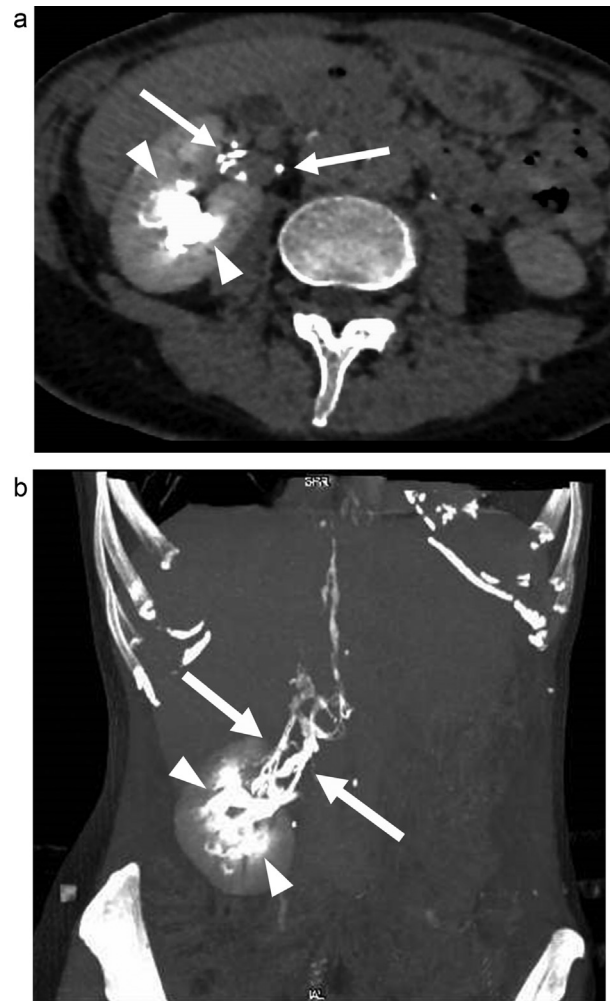


Figure 2 – A delayed phase of contrast-enhanced computed tomography scan (A: axial view, B: coronal view) showed thin lymph vessels (arrows) at the right renal hilum, suggesting communication between the right urinary system and the lymph system. Arrowheads indicate dilated renal calyx and pelvis.

lated to the directional flow of urine from the urinary system to lymph vessels through a uro-lymphatic fistula (Fig. 2 and 3).

Uro-lymphatic fistulas associated with urolithiasis are rarely reported because ureteral stones or hydronephrosis can be usually diagnosed with ultrasonography or plain X-ray examination, but it is difficult to detect fistulas using these modalities [2–4]. In cases in which communication between the renal collecting system and lymphatic system is present concomitant with obstruction of the urinary system secondary to urolithiasis, CT urography—that is, the delayed phase of contrast-enhanced CT—may be useful for diagnosis. In contrast, when lymph vessels are occluded by several disorders, uro-lymphatic fistulas can be diagnosed using lymphangiography [2,5].

Uro-lymphatic fistulas are conservatively managed in most cases [4–6]; however, when clinical symptoms, including severe chyluria, are remarkable, treating the primary diseases

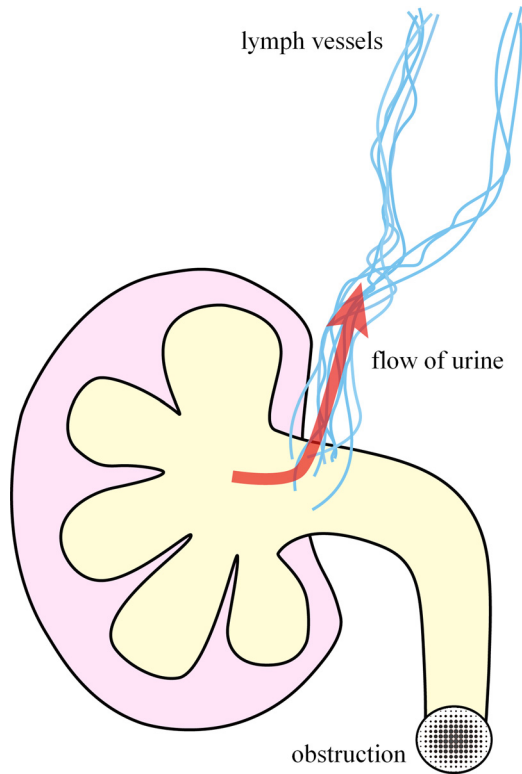


Figure 3 – A schema of uro-lymphatic fistula caused by obstruction of the urinary system. The urine flows into the lymphatic system.

causing the obstruction of lymph vessels or the urinary system may be required [4–6]. As urolithiasis was conservatively managed in this case, no specific treatment for the fistula was needed.

This case report has several limitations. Whether the uro-lymphatic fistula described here was induced by increased pressure in the urinary system due to urolithiasis or the fistula was present prior to the current attack is unknown. Moreover, as the patient's clinical symptoms improved in the short term, a follow-up contrast-enhanced CT imaging study was not obtained. Therefore, whether the present CT finding of the

uro-lymphatic fistula changed after the removal of urolithiasis is unknown.

In summary, we presented the case of a patient with a uro-lymphatic fistula associated with urolithiasis. The fistula was incidentally found on contrast-enhanced CT. Although urine directly flowed into lymph vessels, no significant clinical symptoms were observed. Further studies are required to determine the clinical implication of this disorder.

IRB approval

Institutional review board in the institution approved the publication of this case.

Patient consent

We obtained written and informed consent for the publication of this case from the patient.

REFERENCES

- [1] Stainer V, Jones P, Juliebø SØ, Beck R, Hawary A. Chyluria: what does the clinician need to know? *Ther Adv Urol* 2020;12:1–10.
- [2] Yu NC, Raman SS, Patel M, Barbaric Z. Fistulas of the genitourinary: a radiologic review. *Radiographics* 2004;24:1331–52.
- [3] Rajaonarisona N, Ahmadb A, Cucchia JM, Ortegac JC, Brunetona JN. Reversible uro-lymphatic fistula. *Clin Imaging* 2012;36:72–4.
- [4] Abeygunasekera AM, Sutharshan K, Balagobi B. New developments in chyluria after global programs to eliminate lymphatic filariasis. *Int J Urol* 2017;24:582–8.
- [5] Graziani G, Cucchiari D, Verdesca S, Balzarini L, Montanelli A, Ponticelli C. Chyluria associated with nephrotic-range proteinuria: pathophysiology, clinical picture, and therapeutic options. *Nephron Clin Pract* 2011;119:248–54.
- [6] Kim RJ, Joudi FN. Chyluria after partial nephrectomy: case report and review of the literature. *Sci World J* 2009;9:1–4.