

CASE REPORT

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Sudden fatal bleeding from a uretero-arterial fistula combined with pre-existing uretero-colic and uretero-vaginal fistulas 7 years after a cervical cancer surgery: a case report

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Abstract

Background: Uretero-arterial fistulas (UAFs) are a rare cause of hematuria and could be fatal often due to a diagnostic delay despite recent advances in the treatment modalities.

Case presentation: A 52-year-old woman with a history of advanced cervical cancer developed a fever and was diagnosed with a left uretero-colic fistula. She also had a uretero-vaginal fistula and suffered from repeated urinary tract infections over 6 years. While waiting for an elective colostomy, she developed sudden perineal bleeding and died 14 h after the onset. The autopsy findings revealed that bleeding from a newly developed UAF spreads out to the extracorporeal space through the pre-existing fistulas.

Conclusions: Bleeding from a UAF complicated by other uretero-genital and/or uretero-enteric fistulas could proceed rapidly resulting in a fatal outcome because of a lack of a tamponade effect. Early recognition of a UAF in high-risk patients is crucial for a prompt diagnosis, which might lead to a treatment success.

Keywords: Uretero-arterial fistula, Uretero-genital fistula, Uretero-enteric fistula

Background

Uretero-arterial fistulas (UAFs) are a rare cause of hematuria but could be life-threatening despite the recent advances in the therapeutic modalities. This may be due to the diagnostic difficulty and a lack of recognition of the disease. We report a fatal case of a UAF combined with pre-existing uretero-enteral and genital fistulas that proceeded rapidly.

Case presentation

A 52-year-old woman was referred to the emergency room (ER) for a fever. She had undergone an extended hysterectomy for cervical cancer (stage IIB, pT2aN1M0) 7 years prior and developed a left

uretero-vaginal fistula secondary to postoperative chemoradiotherapy managed with an indwelling ureteral stent (Fig. 1). Following that, she suffered from repeated urinary tract infections occasionally treated by antibiotics and required routine ureteral stent exchanges. She also had type 2 diabetes mellitus treated with pioglitazone hydrochloride over 6 years and revealed no signs of a cancer recurrence during that period. Magnetic resonance imaging revealed a fistula formation between her sigmoid colon and left ureter (Fig. 2). It was considered that a fecal leakage from the uretero-colic fistula worsened the urinary tract infection and an elective colostomy was planned.

One month after the diagnosis of the uretero-colic fistula while waiting for the colostomy, she visited the ER complaining of perineal bleeding. A physical examination revealed hypotension (systolic blood

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pressure 70 mmHg) and tenderness of her lower abdomen but without any bloody stool upon a rectal digital examination nor vaginal bleeding on colposcopy. The laboratory examination revealed an elevated C-reactive protein (CRP) level without anemia and microscopic hematuria and pyuria. She was diagnosed with urosepsis caused by a uretero-colic

fistula and was admitted to the department of gynecology. Although she developed macroscopic hematuria after inserting a urinary catheter, the site of bleeding could not be identified by contrast-enhanced computed tomography (CT). Thirteen hours after the onset, her hypotension worsened resulting in hypovolemic shock (systolic blood pressure 40 mmHg) and massive genital bleeding became evident. She died 1 h later (14 h after the onset) despite intensive care.

The autopsy findings revealed a large amount of blood in the left renal pelvis, bladder, and sigmoid colon. In addition, the left internal iliac artery branch firmly adhered to the ureter and was found to form a UAF, which connected to the uretero-vaginal and uretero-sigmoid fistulas through the ureter (Fig. 3). These findings suggest that bleeding from the newly developed UAF spreads not only into the urinary tract but also over the genital and intestinal tracts through the pre-existing fistulas resulting in sudden death. A histopathological examination of the UAF revealed extensive inflammation with necrosis and an infiltrative squamous cell carcinoma (nonkeratinizing type) in the surrounding tissue (Fig. 4). Besides the previous surgery followed by chemoradiotherapy, her having suffered from repeated infections due to the indwelling ureteral catheter and the occult recurrent cancer tissue might have caused those histological changes that led to the fistula formation.

Discussion

Since first being described in 1908 [1], over 100 reports on UAF have been published to date [2] and its incidence will increase due to more frequent use of indwelling ureteral catheters because of the recent increase in cancer survivors [3]. UAF can occasionally be fatal with a mortality rate of 20–30% even in the modern era [4–6]. Eighty-five percent of UAFs are reported to be a secondary etiology after pelvic surgery or radiation [7], and cervical cancer is one of the most frequent causes of the disease [2, 4]. Our case had several other risk factors for UAF, such as a history of a chronic indwelling ureteral stent, repeated urinary tract infections, and diabetes mellitus as comorbidities. Furthermore, the autopsy confirmed the local recurrence of cervical cancer, which had never been detected during the postoperative surveillance. Those combined risk factors might have caused chronic pressure between the ureter and left internal iliac artery branch resulting in histopathological ischemic changes. In addition, it was considered that the pre-existing uretero-colic and uretero-vaginal fistulas contributed to this sudden

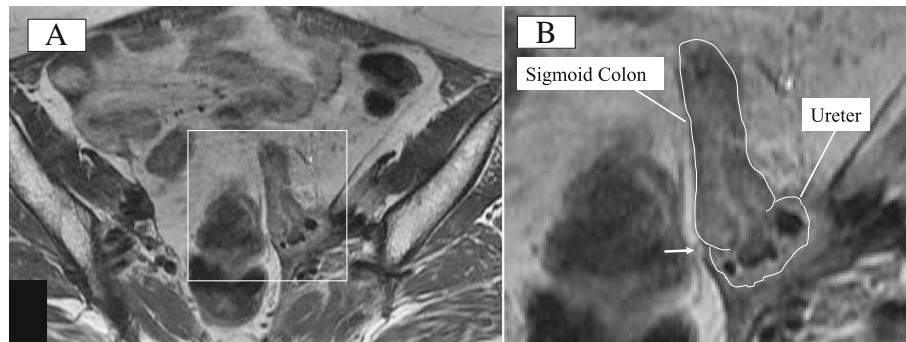


Fig. 2 Magnetic resonance imaging revealed a fistula formation (arrow) between the sigmoid colon and ureter (**a** overall view and **b** enlarged view)

fatal outcome. Because the left ureter had already become connected to the extracorporeal space through these fistulas, bleeding from the newly developed UAF spreads out through that route without a tamponade effect. Therefore, a prompt diagnosis and treatment were quite essential especially in our case.

Endovascular management has been widely used as the main treatment strategy for UAFs over a traditional open repair [8–11] and also has been successfully applied to the treatment of arterio-enteric fistulas [12, 13]. It would be promising to enable immediate hemostasis with less morbidity than surgical treatment, especially in patients who had prior pelvic interventions and a vascular pathology.

Despite these recent advances in the treatment modalities, diagnosing UAF is still a clinical challenge, and a

diagnostic delay has been reported to be associated with a significantly poor outcome [14–16].

Although contrast CT, cystoscopy, and standard angiography have been employed as diagnostic modalities, the clinical utility or sensitivity are far from satisfactory and the diagnosis is often made at the time of surgery [2, 15, 17]. In addition to the lack of awareness of the etiology, even the most sensitive angiography could often fail to visualize extravasation into the ureter due to intermittent bleeding and the presence of an indwelling ureteral stent [2, 18, 19]. To improve the diagnostic accuracy, pharmacological provocative maneuvers other than standard angiography have been provocative angiography in which the indwelling ureteral stent is removed to help visualize the fistula [6, 14, 18, 20]. Contrast-enhanced CT was not diagnostic even after the onset of the bleeding in our case and therefore angiography including provocative maneuvers might have helped obtain a correct diagnosis. However, because stent manipulation not only can be diagnostic but also cause massive bleeding [21, 22], provocative angiography should be attempted with simultaneous arterial embolization or surgical bypass grafting [6]. Further, it was considered that to suspect a UAF by itself was a key issue. Considering the history of pelvic interventions including chemoradiotherapy and chronic indwelling ureteral catheters requiring routine exchanges due to repeated urinary tract infections, we should have suspected the possibility of the UAF during the earlier stage of the workup in our case.

Conclusions

Bleeding from UAFs complicated with other uretero-genital and/or uretero-enteric fistulas could proceed rapidly resulting in a fatal outcome. It was considered that both a diagnostic delay and concomitant fistula, which eliminated the tamponade effect against bleeding, were responsible for the sudden and

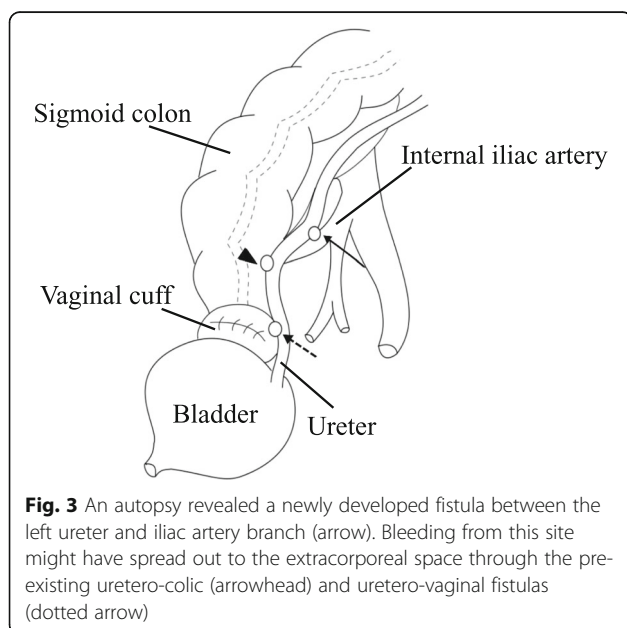


Fig. 3 An autopsy revealed a newly developed fistula between the left ureter and iliac artery branch (arrow). Bleeding from this site might have spread out to the extracorporeal space through the pre-existing uretero-colic (arrowhead) and uretero-vaginal fistulas (dotted arrow)

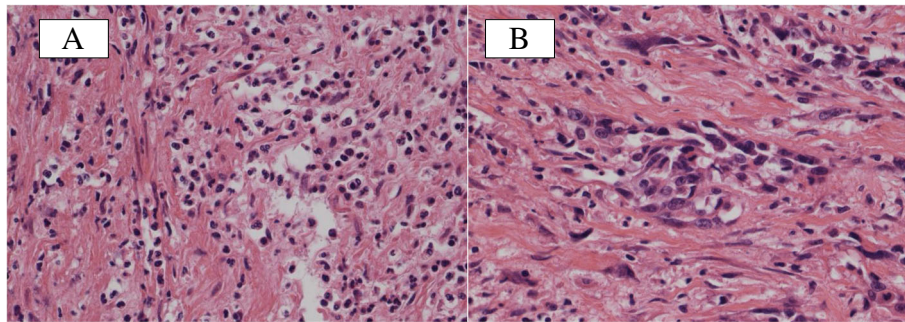


Fig. 4 A histopathological examination revealed extensive inflammation and necrosis around the uretero-arterial fistula (a hematoxylin eosin staining) and an infiltrative squamous cell carcinoma in the surrounding tissue (b hematoxylin eosin staining)

fatal outcome in our case. Moreover, we should be aware of UAFs as the most critical cause of hematuria in high-risk patients and early recognition of the disease itself is a key issue both for the diagnostic and a successful treatment especially in cases with other ureteral fistulas.

Abbreviations

CRP: C-reactive protein; CT: Computed tomography; ER: Emergency room; UAF: Uretero-arterial fistula

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Authors' contributions

HY wrote this paper. HA and YO performed the autopsy, reviewed the pathological findings, and revised the manuscript. MT reviewed the medical image findings. TN helped to write the manuscript. All authors read and approved the final manuscript.

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Consent for publication

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