Uretero-aortic Fistula: A Case Report

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We report a case of uretero-aortic fistula following prolonged ureteral stenting in the left ureter which crossed over the aorta toward the cutaneous ureterostomy stoma. A 59 year-old woman presented massive bleeding from the left cutaneous ureterostomy of the single stoma for bilateral ureters. The patient underwent radical hysterectomy and total cystectomy with a single stoma cutaneous ureterostomy for advanced cervical and bladder cancers. The postoperative course was uneventful except for pyelonephritis due to bilateral ureteral stenosis, which were treated by bilateral ureteral stenting. The patient had intermittent hematuria from the left cutaneous ureterostomy for 4 years after ureteral stenting. Massive bleeding from the left cutaneous ureterostomy requiring a big amount of blood transfusion followed intermittent hematuria, finally. A Computed tomography (CT) suggested both severe adhesion of the left ureter to the aorta and left renal pelvic hematoma. Massive bleeding seemed to be caused by uretero-aortic fistula. A fistula, about 5 mm in diameter, from the left ureter to the aorta was detected at exploration. The defect of aortic wall was closed primarily and then left nephroureterectomy underwent for nonfunctioning kidney. Her postoperative course was uneventful at the 18-month follow-up.

Key words: uretero-aortic fistula, ureteral stenting, single stoma cutaneous ureterostomy

INTRODUCTION

Uretero-aortic fistula, one of ureteroarterial fistulae, is a rare clinical occurrence [1–3]. Adequate diagnosis and critical care management are crucial to rescue the patient from this life-threatening condition. Patients with a history of radical pelvic surgery, ureteral stenting, and prior pelvic radiation therapy seem to have greater risk of ureteroarterial fistula. While the mortality rate was 70% before 1980, it decreased to about 14% recently because of improvement of critical care management [1]. We should be aware that ureteroarterial fistula can occur as a serious complication of prolonged ureteral stent indwelling.

CASE REPORT

A 59 year-old woman presented massive hemorrhage from the left cutaneous ureterostomy of the single stoma for bilateral ureters. The patient underwent radical hysterectomy and total cystectomy with a single stoma cutaneous ureterostomy for advanced cervical and bladder cancers after chemotherapy for cervical cancer. The postoperative course was uneventful except for pyelonephritis due to bilateral ureteral stenosis, which were treated by bilateral ureteral stenting. The patient had intermittent hematuria from the left cutaneous ureterostomy for 4 years after ureteral stenting. Massive bleeding from the left cutaneous ureterostomy requiring a big amount of blood transfusion followed intermittent hematuria, finally.

A Computed tomography (CT) (Fig. 1A) showed dilated left renal pelvis with hemorrhage in it. However it also suggested adhesion at the crossing point of the left ureter over the aorta (Fig. 1B), uretero-aortic fistula was not clearly detected. Intermittent but self-limiting massive bleeding followed the first massive bleeding in the ensuing week. Although even a Magnetic resonance (MR) angiography (Fig. 2) could not reveal an uretero-aortic fistula, massive bleeding was suspected from the fistula between the left ureter and the aorta. After further counseling with the family, we performed an exploration. A fistula, about 5 mm in diameter, from the left ureter to the aorta was detected at exploration. The defect of aortic wall was closed primarily and then left nephroureterectomy underwent for nonfunctioning kidney. Her postoperative course was uneventful at the 18-month follow-up.

DISCUSSION

Although fistula formation between the urinary tract and the arterial system (UAF) is a rare clinical occurrence [1–3], adequate diagnosis and critical care management are crucial to rescue the patient from this life-threatening condition.

UAF include ureteroarterial fistula [1–3], aortoureteric fistula [4–6], arterio-ureteric fistula [7], and iliac artery-to-ureteral fistula [8]. Naming of those UAF is made in order of primary disease organ's name first and then targeted organ's name. UAF can occur in association with prolonged ureteral stenting, pelvic surgery, previous radiation therapy, vascular disease, and vascular pathology. One or more of these five specific risk factors contributed to the formation of UAF in all reported cases [3]. Pelvic surgery and ureteral stenting were the most frequently cited risk factors. Prolonged ureteral stenting was supposed to be the cause of UAF

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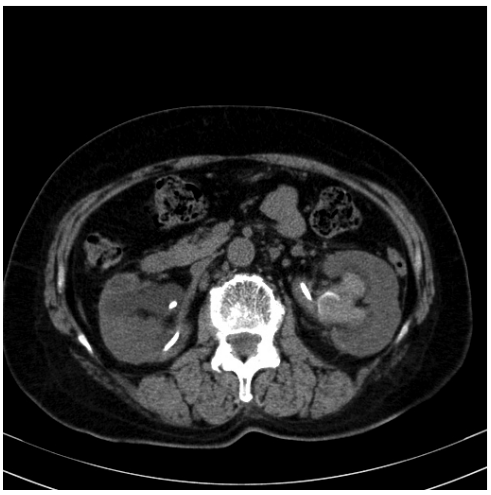

Fig. 1A A CT showed dilated left renal pelvis with hemorrhage in it.

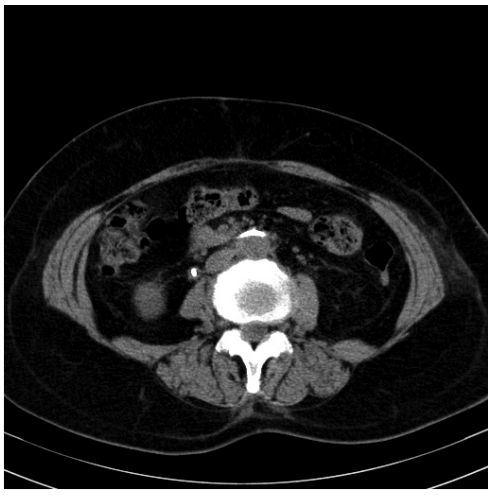

Fig. 1B Although a CT suggested adhesion at the crossing point of the left ureter over the aorta, uretero-aortic fistula was not detected.

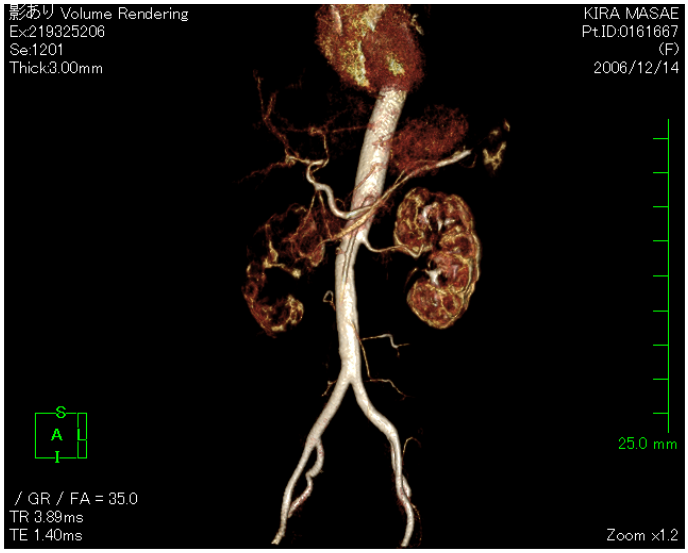

Fig. 2 A MR angiography showed neither aneurysm nor fistula.

in this case, so that we called this case as uretero-aortic fistula.

Presentations of UAF range from microscopic hematuria or intermittent hematuria to life-threatening bleeding. UAF is so rare that physicians often select to treat patients with other diagnosis, such as urolithasis. Even if physicians entertain the diagnosis of UAF, no evidence of UAF was seen in the majority cases [3]. Routine workup studies didn't show any evidence of UAF also in our case. Once UAF is identified or suspected, prompt planning with radiologists, urologists, and vascular surgeons must be undertaken. Multiple options are available for treatment of UAF. Treatment must include repair of the artery and ureter. The vascular defect can be repaired using an open approach, with embolization, ligation, and extra-anatomic arterial reconstruction, or with endovascular stenting [6]. Due to improved critical care management, the mortality rate decreased from about 70% to 14%. Stephen *et al.* [3] reported that management of the arterial component of the fistula is affected by: (1) associated local infection, (2) the presence of the associated aneurismal or occlusive disease in the iliac and "downstream" arteries, and (3) the available collateral circulation to the ipsilateral leg. Most commonly applied procedure is open surgical approaches (*i.e.* simple ligation of the involved artery with or without bypass reconstruction or direct suture) with concomitant urinary diversion. Keller *et al.* [8] reported a successful use of vasoocclusive technique to treat UAF. Disadvantage of this technique required reconstruction procedure. Recently Inoue *et al.* [9] reported a successful intraluminal ureteral occlusion technique to manage UAF with long term control. They said that the uroepithelial hyperplasia together with thrombus caused by coils might play some roles. In our case, adhesion of the left ureter to the aorta was not so severe that we performed a stump closure of the aorta and following left nephroureterectomy. Although preservation of the kidney has priority, if possible, from urologic perspective, nephrectomy

was performed in about 50% of UAF cases but resulted in the best outcome for the patients with normal physical and social function [3].

UAF is a rare yet life-threatening condition, so every physician should attempt to prevent its formation. The occurrence of UAF with prolonged ureteral stenting after pelvic surgery or urinary diversion should be avoided by careful surgery and postoperative management.

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