



Endovascular Treatment for Arterioureteral Fistula of the Abdominal Aorta: A Case Report and Literature Review

복부 대동맥에 발생한 동맥-요관 누공의 혈관 내 치료: 증례 보고와 문헌고찰

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We present a rare case demonstrating successful endovascular management of an arterioureteral fistula involving the abdominal aorta. Arterioureteral fistulas are rare but life-threatening, with mortality rates ranging from 7% to 23%. Early recognition and prompt management are essential for preventing catastrophic consequences, including hypovolemic shock. However, recognition of an arterioureteral fistula requires a high index of clinical suspicion due to its rarity and the lack of a sensitive diagnostic method. Arterioureteral fistulas could be induced by traumatic events in patients who have a history of pelvic surgery, radiation, and prolonged placement of a ureteral stent. Endovascular stent graft placement could be a valid treatment option for arterioureteral fistulas involving the abdominal aorta.

Index terms Urinary Fistula; Hematuria; Stents; Endovascular Procedures; Aneurysm, False

INTRODUCTION

The arterioureteral fistula (AUF) is a rare abnormal communication between the artery and ureter. Since the first description of AUF in 1908 reported after bilateral ureterolithotomies, the incidence of this condition is increasing (1). The mortality rate for patients with an undiagnosed AUF is reported up to 52% and early recognition and prompt management are essential for preventing catastrophic consequences (2, 3). However, AUF remains a diagnostic challenge to most physicians due to its rarity and lack of sensitive diagnostic method (4). Herein, we present a case of AUF involving ab-

Received May 22, 2019
 Revised September 22, 2019
 Accepted September 27, 2019

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dominal aorta successfully treated with endovascular stent graft placement.

CASE REPORT

A 62-year-old female was admitted for gross hematuria from both ileal conduit and left nephrostomy tube after slip-down. Twenty months before, the patient underwent pelvic exenterating with ileal conduit, radiation and ureteral stent placement for recurrent cervical cancer. Vital signs were as follows: blood pressure 100/60, heart rate 96/min, respiratory rate 18/min and temperature 36.4°C. Laboratory studies revealed a hemoglobin level of 7.0 g/dL and hematocrit level of 20.7%. Abdomen CT scan revealed small amount of hematoma in the left renal pelvis without evidence of active bleeding, suggesting possible renal injury. Two units of packed red blood cells were transfused. However, gross hematuria continued and significant drop of hemoglobin was noted on the following day; hemoglobin level of 6.3 g/dL and hematocrit level of 18.9%.

The patient was referred to the interventional radiology for diagnostic angiography and further therapeutic embolization if needed. Selective both renal angiography using a 5-F catheter (Yashiro; Cook Medical, Bloomington, IN, USA) showed no active bleeding focus. Subsequent aortography using a 5-F pigtail catheter (Cook Medical) revealed small pseudoaneurysm at distal abdominal aorta and fistula formation with adjacent left ureter, suggesting AUF (Fig. 1A, B). A retrospective review of abdomen CT scans identified corresponding minimal irregularity at the level of ureteroarterial crossing (Fig. 1C-E).

Superselective embolization of fistula tract using the glue-lipiodol mixture at a 1:2 ratio was attempted, but post-embolization angiography showed persistent flow into ureter. As a secondary strategy, an endovascular stent graft treatment was adopted as a lifesaving procedure. A 23 mm × 49 mm stent graft (Endurant IIs; Medtronic Vascular, Santa Rosa, CA, USA) was deployed at the distal abdominal aorta covering the opening of fistula. Following angiography revealed complete exclusion of pseudoaneurysm and immediate hemostasis can be achieved (Fig. 1B). After endovascular treatment, hematuria disappeared and hemoglobin level increased to normal levels. Broad spectrum antimicrobial prophylaxis with Piperacillin-tazobactam and oral antiplatelet therapy with aspirin were administered. The patient was eventually discharged in stable condition without significant complication after three weeks.

DISCUSSION

The pathophysiology of AUF is still not well understood, but inflammatory or ischemic injuries to the ureters and adjacent vascular structures has been suggested. Previous surgery and radiation could induce fibrotic inflammatory process, which fixes the ureter to the anterior wall of adjacent artery (5). Ischemic vascular injury induced by radiation damage and pressure necrosis may eventually result in fistula formation. The prolonged ureteral stent placement may also attribute to the additional injury of an already compromised ureter.

The typical location of AUF is at the ureteroarterial crossing at the pelvic brim, which is compatible with presumed pathophysiology. According to the previous systematic review, 63% of fistulas involved the left ureter and iliac artery (6). The AUF involving the aorta is extremely

Fig. 1. A 62-year-old female with gross hematuria from both the ileal conduit and left nephrostomy tube after a slip-down.

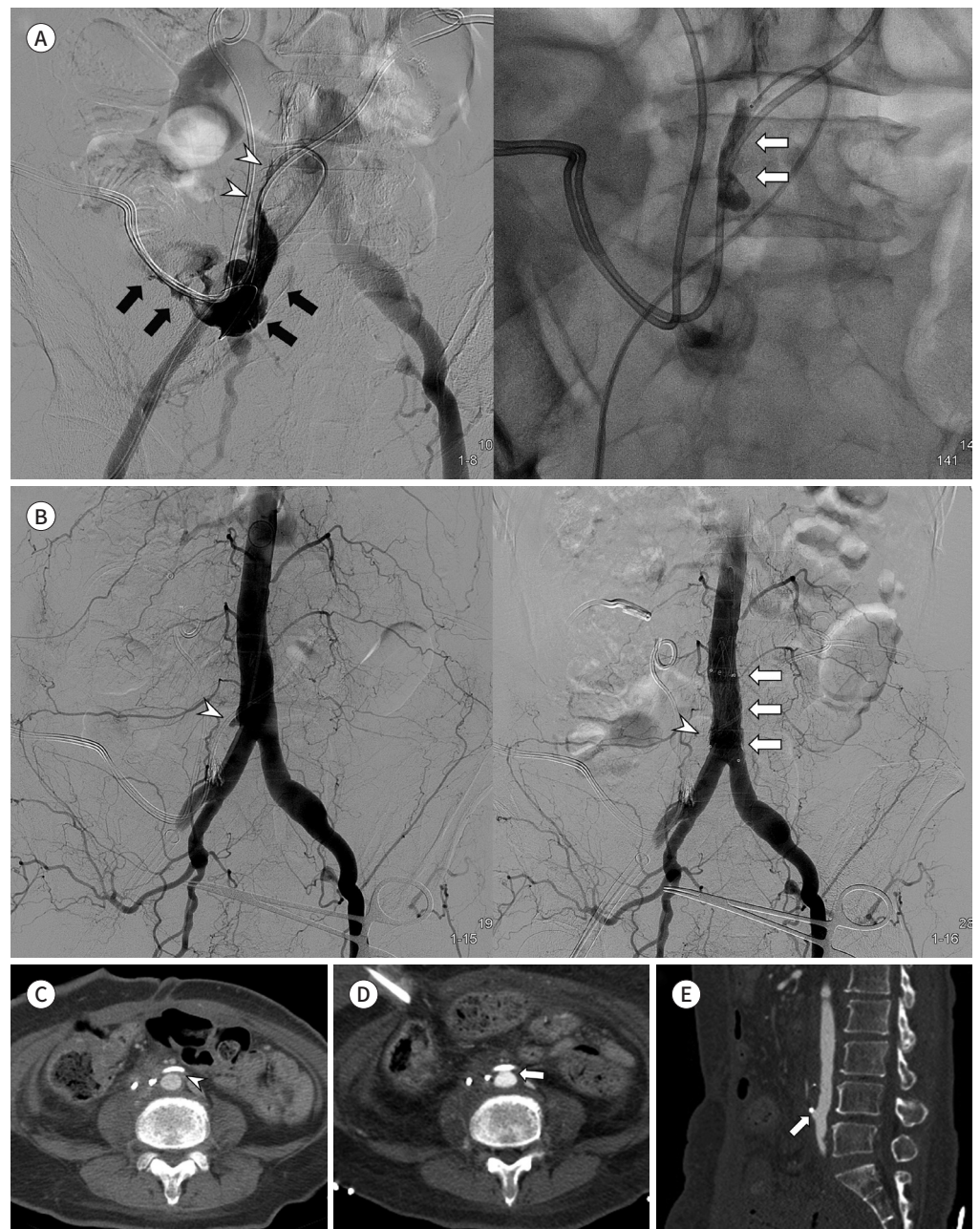
A. Aortography (left image) shows an arterioureteral fistula (arrowheads) and active contrast extravasation (arrows) along the left ureteral stent and ileal conduit. Superselective embolization of the fistula tract (right image) was performed using a glue-lipiodol mixture (arrows).

B. Aortography after glue embolization (left image) shows a small remnant fistula (arrowhead). A 23 mm × 49 mm self-expandable stent graft (right image, arrows) was deployed at the distal abdominal aorta with complete exclusion of the fistula (arrowhead).

C. Contrast-enhanced abdominal CT scan obtained three months before the slip-down demonstrates the intact anterior wall of the aorta (arrowhead) and an overlying ureteral stent.

D. Contrast-enhanced abdominal CT scan on admission shows subtle irregularity of the anterior wall of the aorta (arrow) at the level of the ureteroarterial crossing.

E. Sagittal reformatted contrast-enhanced CT scan reveals a corresponding small outpouching lesion (arrow) at the distal abdominal aorta, indicating a pseudoaneurysm.



rare condition, which reported in only 7 patients. In patients with urinary diversion, ureteroarterial crossing takes place at a higher level than usual anatomy. Therefore, the arterial part of fistula could involve the proximal common iliac artery or even the distal abdominal aorta.

The AUF has been classified into primary and secondary types on the basis of etiology (4, 5). The primary fistulas (15%) are related to the vascular abnormalities such as aneurysms, arteriovenous malformations, or aberrant vessels. The secondary fistulas (85%) have been reported after prior pelvic interventions including pelvic surgery, combined with irradiation and with ureteral stent placement. Post-traumatic fistula is extremely rare subgroup with only one case report which describes a patient with a gunshot wound in the abdomen (7).

Gross hematuria is the dominating symptom of AUF, which can be intermittent or life-threatening massive with hypotension. Most patients experienced the first episode of hematuria spontaneously and some patients during the change or insertion of ureteral stent (6). However, hematuria provoked after blunt trauma has not been described before. There is no established explanation how blunt trauma affected AUF. The impact of trauma may disrupt already damaged ureter and arterial wall, promoting the formation of fistula. In the present report, recent trauma history and nonspecific CT finding raise a clinical suspicion of blunt renal injury and result in diagnostic delay. If noninvasive imaging workup failed to identify the cause of hematuria, diagnostic angiography can be helpful in patients with typical clinical triad of pelvic surgery, radiation and ureteral stenting (4).

Cross sectional imaging has a limited role in diagnosis of AUF, because of its low sensitivity. Enhanced CT scan usually negative and the fistula tract is almost never identified. In a small subset of patients with pseudoaneurysm, enhancing lesion could be identified near the ureteroarterial crossing. In the present case, it was difficult to recognize the subtle irregularity of aorta without retrospective careful review. The correct diagnosis could be made after conventional angiography. Digital subtraction angiography remains the primary diagnostic tool, but its sensitivity is less than 50% (5, 8). Angiographic findings include direct extravasation into fistula, pseudoaneurysm and subtle irregularity or intimal defect (4). In cases of negative study, a provocative maneuver such as manipulation of ureteral stent could help visualization of extravasation (8). However, balloon tamponade should be prepared for subsequent massive hemorrhage.

A wide variety of treatment options have been suggested in the literature (6). Because the majority of fistulas are secondary to previous pelvic surgery and radiotherapy, open repair is often not feasible (9, 10). Currently, endovascular stent graft placement has become the treatment of choice for AUF with less morbidity and mortality (4, 6). Long-term follow-up data after stent graft placement is limited due to its rare incidence. Previous study with mean follow-up of 15.5 months revealed possible complications including recurrent bleeding, lower extremity complications and stent graft complications, and recommended the use of antibiotics and long-term anticoagulant therapy (10).

In conclusion, recognition of AUF requires a high index of clinical suspicion due to its rarity and lack of sensitive diagnostic method. It could be provoked after traumatic event in patients who have a history of pelvic surgery, radiation and prolonged placement of ureteral stent. The endovascular stent graft treatment could be a valid treatment option for AUF involving abdominal aorta.

Author Contributions

Writing—original draft, all authors; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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복부 대동맥에 발생한 동맥-요관 누공의 혈관 내 치료: 증례 보고와 문헌고찰

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저자들은 복부 대동맥에 발생한 동맥-요관루를 혈관 내 접근을 통해 성공적으로 치료할 수 있었던 드문 증례를 보고하는 바이다. 동맥-요관루는 극히 드물지만, 사망률이 7~23%에 이르는 치명적인 질환이다. 저혈량쇼크와 같은 치명적 합병증을 예방하기 위해서는 조기 진단과 함께 즉각적인 치료가 필수적이다. 하지만 질환 자체가 희귀하고 민감도가 높은 검사 방법이 없기 때문에, 진단을 위해서는 높은 수준의 임상적 의심이 반드시 필요하다. 복강 내 수술, 방사선 치료 및 요관 스텐트의 장기 설치 등의 특징적인 과거력을 가진 환자에서 외상적 사건이 동맥-요관루 발생의 촉발 요인이 될 수 있다. 복부 대동맥에 발생한 동맥-요관루 환자에서도 혈관 내 인조혈관 스텐트의 삽입은 효과적인 치료 방법이다.

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