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Endovascular treatment of arterio-ureteral fistulae with covered stents: Case series and review of the literature

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Abstract

Arterio-ureteral fistulae are abnormal connections between an artery and the ureter and carry a high mortality. We present two cases of arterio-ureteral fistulae that presented with life-threatening hematuria. Both patients were treated with endovascular covered stent placement.

Keywords

Arterio-ureteral fistula, vasculoureteric fistula, hematuria

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Introduction

Arterio-ureteral fistulae (AUFs) are abnormal connections between blood vessels and the ureter and most commonly involve the iliac arteries and the ureter. They are rare entities, and only around 140 cases have been reported in the literature.[1] Primary fistulae may occur due to aorto-iliac atherosclerotic disease. However, the majority of fistulae are secondary, occurring due to radiation or surgery for urological and gynecological cancers.[2] The incidence of AUFs is reportedly increasing, which may be due to the increased use of ureteral stents, higher dosage used for radiation therapies, and increasing numbers of vascular and pelvic surgeries.[2,3] We present two cases of AUFs involving the iliac arteries, related to gynecologic malignancies and therapies related to them. Both patients were treated successfully with endovascular covered stent placement.

Our procedure consents include a statement regarding the use of images such as radiographs without patient identifiers for teaching and illustrative purposes. Our institutional policy does not require patient consents for case reports. Case reports are also exempt from institutional board review.

Case 1

A 44-year-old female was transferred to our emergency department from an outside institution for the management of sudden onset, profuse vaginal bleeding and hematuria. Her history was significant for cervical cancer diagnosed 3 years prior to presentation, and she was post total abdominal hysterectomy and pelvic lymphadenectomy, as well as multiple chemotherapy and radiation therapy regimens. Her history was further complicated by persistent bilateral hydroureter and hydronephrosis for 6 months prior to presentation, likely secondary to radiation ureteritis.

On arrival, the patient was in profound hemorrhagic shock and diaphoretic with active vaginal bleeding. Her blood pressure was 74/49 mmHg, pulse rate was 117 beats/min, and respirations were 20/min. A Foley catheter was placed in the bladder, which yielded fresh blood and clots. She was taken emergently to the operating room by the gynecological surgery team for exploration and vaginal packing. Her bleeding seemed to respond initially after vaginal packing; however, profuse bleeding was subsequently noted in the operating room. At this time, the decision was made to take the patient from the operating room to the interventional radiology suite for an angiogram.

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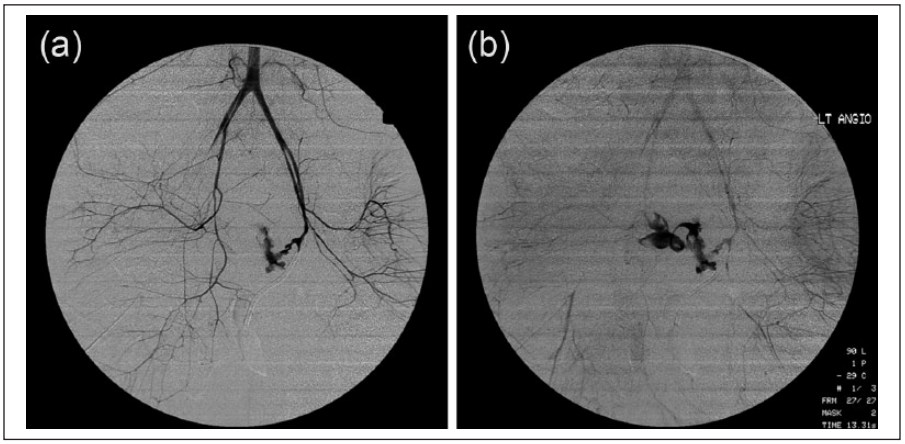

Figure 1. (a) Aortogram shows extravasation from the left external iliac artery and (b) delayed images during the angiogram show pooling of contrast in the pelvis centrally.

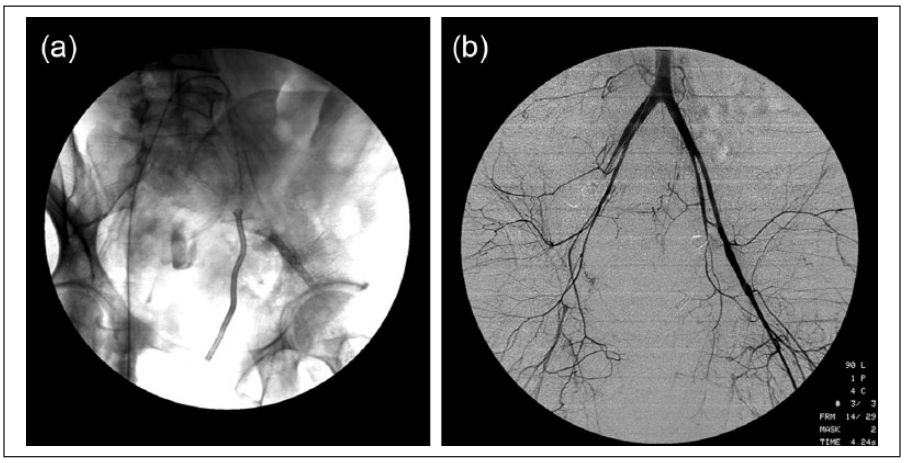

Figure 2. (a) Fluoroscopic spot image during placement of a 7 mm × 30 mm Wallgraft covered stent (arrow) and (b) aortogram after covered stent placement in the left external iliac artery demonstrates no extravasation.

A pelvic aortogram showed active extravasation of contrast from the left external iliac artery into the pelvis, in proximity to the course of the left ureter (Figure 1(a)). Contrast material appeared to pool centrally in the pelvis toward the vagina (Figure 1(b)). Given the finding of extensive vaginal bleeding along with hematuria, the diagnosis of arterio-ureteral-vaginal fistula was considered. A 7 mm × 30 mm covered stent (Wallgraft Endoprosthesis; Boston Scientific, Natick, MA, USA) was then placed across the area of extravasation from the left external iliac artery (Figure 2(a)). A repeat angiogram demonstrated no further extravasation of contrast from the previous site of hemorrhage (Figure 2(b)). The patient gradually became hemodynamically stable and was observed in the intensive care unit. Hematuria and vaginal bleeding resolved over the next 4 days. She was discharged 16 days later in stable condition without further episodes of hematuria or vaginal bleeding. The patient did not have any recurrence of her fistula for 12 months, after which she was lost to follow-up.

Case 2

A 71-year-old female with a history of metastatic cervical cancer treated with surgical debulking, chemotherapy, and radiation presented to our emergency room with hematuria. She had a history of a right ureteric stent secondary to distal obstruction by her pelvic mass. Six days prior to presentation, she had a right nephrostomy placement due to recurrent hydronephrosis and suspected stent obstruction. On presentation, she was hypotensive and tachycardic with blood in her nephrostomy and Foley catheter bag. Vascular injury during her nephrostomy placement was suspected, and a renal arteriogram was performed (Figure 3(a)). The renal arteriogram showed no evidence of extravasation or arterial injury, and no interventions were done. She was managed with observation and transfusions. Her hematuria did not clear in next 3 days, and a renal angiogram was repeated, which again was negative for source of bleed. An embolization of the lower pole artery in the vicinity of the catheter

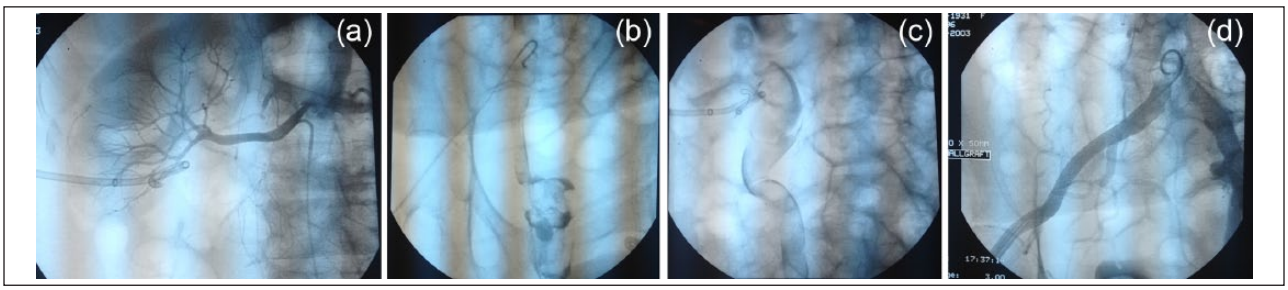

Figure 3. (a) Fluoroscopic spot image from a right renal arteriogram shows no evidence of vascular injury, (b) fluoroscopic spot image during injection of the right common iliac artery shows contrast entering into the ureter, (c) contrast noted refluxing up into a dilated proximal ureter after injection of the right common iliac artery, and (d) aortogram after placement of a covered stent in the right common iliac artery demonstrates exclusion of the arterio-ureteral fistula.

was then done using a single 5 mm Nestor coil (Cook, Bloomington, IN, USA). After the embolization, the catheter was withdrawn to the right common iliac artery and an angiogram was performed which outlined the ureter (Figure 3(b)), with contrast reflux noted up the dilated right ureter to the level of the kidney (Figure 3(c)). A diagnosis of an AUF was made. A 10 mm × 50 mm covered stent (Wallgraft Endoprosthesis; Boston Scientific) was then placed across the fistula. Aortogram after placement of the covered stent showed exclusion of the AUF (Figure 3(d)). The patient's hematuria resolved over the next 5 days, and she was discharged home 7 days later. She did not have any recurrent episodes of hematuria and subsequently succumbed to her malignancy 3 months after the episode.

Discussion

AUF is a rare but potentially life-threatening entity. Primary fistulae, which account for about 15% of cases, emanate from aortic aneurysms or vascular malformations eroding into the ureter.[2,4] Secondary fistulae are more common and account for 85% of cases and are most often seen in patients with previous pelvic cancer surgery.[2] Other causes include vascular surgery involving common or external iliac arteries, as well as urinary diversion surgery and urinary stenting. Van den Bergh et al.[1] reviewed 139 published cases of AUFs and found that 54% of the patients had previous cancer, 31% had prior vascular surgery, 37% had received radiotherapy, 61% had ureteral stenting, and 34% had ureteral deviation. Bergqvist et al.[2] also found that the majority of patients with secondary AUFs had a complicated course after their original pelvic or vascular surgery. Both our cases had a history of pelvic malignancy, surgery, and radiotherapy.

The initial clinical presentation of AUF is hematuria, the severity of which can vary from minimal bleeding to that of life-threatening exsanguination.[1–3,5] Other symptoms include back pain, urinary tract infection, and urinary retention. The presence of an aneurysm or pseudoaneurysm involving the iliac vessels has been reported in up to 38% of patients.[1] Diagnosis of AUFs can often be challenging. Conventional angiography may be used for diagnosis as well as subsequent surgical or endovascular treatment planning. A negative angiogram, however, does not exclude the diagnosis as angiograms have been reported to be positive in only 69% of cases.[1] Ureteral contrast studies that are positive in 52% cases are the next best diagnostic modality.[1] Computed tomography (CT) urography may show contrast in ureters, but often the miniscule nature of fistula makes this an infrequent finding.[4] Provocative maneuvers, such as advancing and withdrawing an angioplasty balloon catheter around the suspected region, can help with the diagnosis.[6] However, it is recommended to keep at hand a balloon mounted on a ureteral wire and placed in the offending vessel, bladder, or ileal conduit just below the ureter, which can be advanced into fistula when the bleeding is provoked.[4] Cystoscopy may also be used for diagnosis and shows pulsatile bleeding at the site of the fistula.[5]

Currently, overall mortality varies from 7% to 23%. Prompt diagnosis and treatment are key, and mortality as high as 58% has been reported in patients in whom diagnosis is delayed.[4,7] Many different surgical and endovascular treatments have been described for the treatment of AUFs. These include arterial ligation, bypass grafting, direct artery repair, ureterectomy, percutaneous nephrostomy, ligation/removal of ureter, nephrectomy and nephroureterectomy, coiling, or stenting of ureters. Since most patients presenting with AUFs have a complicated history of pelvic surgery and/or malignancy, surgery is often not feasible.[4] Endovascular treatment with arterial coil embolization has been described in patients with internal iliac artery involvement. However, since most of the AUFs involve common or external iliac artery, coil embolization is usually not considered.[4] Endovascular treatment of AUFs with covered stents has also been previously described.[1,8] Endovascular treatment with stents is less invasive and provides rapid control of life-threatening bleeding. However, with the use of covered stents, there is a theoretical concern of graft infection due to contamination by bacteria in the urinary tract. However, this does not seem to be an issue in the majority of the reported cases, although the follow-up periods are limited.[4] Our two patients did not have

morbidity related to stent infection. We had no perioperative mortality and no immediate postoperative bacteremia in our cases. Furthermore, follow-up over 12 and 3 months, respectively, showed no evidence of lower limb vascular compromise, loss of stent patency, or reformation of fistula. While our patients had many comorbid conditions, we were able to achieve rapid and successful control of acute hemorrhage in these patients.

Outcomes after surgical or endovascular treatment of AUFs have varied. In their systematic review of treatment for AUFs, Van den Bergh et al.[3] found no fistula-related mortalities in the patients treated with endovascular stent grafts. However, 3 out of 32 patients had thrombosis of the stent and 2 patients experienced recurrent hematuria. In their review, Van den Bergh et al.[1] found that 36 out of 139 patients (26%) died during follow-up after various procedures for the treatment of AUFs. Only 18 of them (13%) died due to recurrence of the fistula. Patients who had developed fistula due to oncological reasons had 13% risk of dying, while those whose fistula was due to vascular reasons had 9% risk of death. Bergqvist et al.[2] also conducted a systematic review of various treatment modalities for AUFs, including endovascular covered stent-graft placement. Of the 80 patients included in their review, the majority of them (53%) had AUFs secondary to pelvic surgery with subsequent radiation therapy. A total of 57 patients underwent interventions, with surgical ligation, nephroureterectomy, or covered stent placement. In patients in whom there was a preoperative diagnosis of AUF, the authors reported 0% mortality. On the other hand, patients without a preoperative suspicion of AUF had a 39% mortality rate. A total of five patients were treated with covered stent placement. Of those five patients, hematuria ceased in initial follow-up in four patients. One patient (20%) had fistula recurrence. Follow-up for these patients was limited, and the authors did not provide mortality data specific to endovascular covered stent-graft repair of AUFs. However, the authors concluded that endovascular covered stent-graft repair of AUFs is a viable short-term treatment option.

Stent-graft treatment of potentially infected sites remains controversial. Communication with the urinary tract certainly introduces a source of infection. However, as in our cases, patients with AUFs often present with life-threatening hemorrhage requiring urgent treatment. Furthermore, patients who present with AUFs often have complicated oncological and surgical histories, increasing the risk of postoperative morbidity and mortality associated with surgical intervention.

Conclusion

In conclusion, endovascular placement of covered stents is a feasible therapeutic option for the treatment of acute life-threatening hemorrhage due to AUFs.

Declaration of conflicting interests

None of the authors have any conflicts of interest.

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