CLINICAL INVESTIGATION

# Long-term Results of Endovascular Stent Graft Placement of Ureteroarterial Fistula

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#### Abstract

*Purpose* To evaluate the safety, efficacy, and long-term results of endovascular stent graft placement for ureteroarterial fistula (UAF).

*Methods* We retrospectively analyzed stent graft placement for UAF performed at our institution from 2004 to 2012. Fistula location was assessed by contrast-enhanced computed tomography (CT) and angiography, and freedom from hematuria recurrence and mortality rates were estimated.

*Results* Stent graft placement for 11 UAFs was performed (4 men, mean age  $72.8 \pm 11.6$  years). Some risk factors were present, including long-term ureteral stenting in 10 (91 %), pelvic surgery in 8 (73 %), and pelvic

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K. Idoguchi e-mail: idoguchi@ares.eonet.ne.jp radiation in 5 (45 %). Contrast-enhanced CT and/or angiography revealed fistula or encasement of the artery in 6 cases (55 %). In the remaining 5 (45 %), angiography revealed no abnormality, and the suspected fistula site was at the crossing area between urinary tract and artery. All procedures were successful. However, one patient died of urosepsis 37 days after the procedure. At a mean follow-up of 548 (range 35–1,386) days, 4 patients (36 %) had recurrent hematuria, and two of them underwent additional treatment with secondary stent graft placement and surgical reconstruction. The hematuria recurrence-free rates at 1 and 2 years were 76.2 and 40.6 %, respectively. The freedom from UAF-related and overall mortality rates at 2 years were 85.7 and 54.9 %, respectively.

*Conclusion* Endovascular stent graft placement for UAF is a safe and effective method to manage acute events. However, the hematuria recurrence rate remains high. A

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further study of long-term results in larger number of patients is necessary.

**Keywords** Endovascular treatment · Long-term results · Stent graft · Ureteral-iliac artery fistula · Ureteroarterial fistula

# Introduction

Ureteroarterial fistula (UAF) is a direct fistulous communication between the ureter and the artery that results in bleeding in the ureter [1]. It is uncommon, but recently the reported cases have been increasing [2–4]. Chronic ureteral stenting, pelvic external radiation, extensive pelvic surgery, and peripheral vascular diseases are known as the main risk factors [5, 6]. The increasing number of UAF cases reflects the increase in the combination of these risk factors, relating to the improved cancer survival and aggressive multimodal treatment for abdominopelvic cancers [6, 7].

Conventional management of UAF would be a direct surgical intervention including local reconstruction or ligation of the iliac artery with or without extra-anatomical bypass. However, the surgical field is often difficult as a result of the previous surgery, radiotherapy, and the presence of residual malignancy, and comorbidities may further increase surgical risk [1, 3, 4, 8, 9]. Stent graft placement for endovascular repair of UAF, which was first reported in 1996 [10] and thereafter became widely known, has multiple advantages, including minimal invasiveness and immediate control of bleeding. Several case reports described that it has good outcomes and potentially reduces perioperative morbidity and mortality [9, 11–18]; however, long-term outcomes are still unknown.

Herein we report our experiences in UAF treatment using endovascular stent graft placement, and we evaluate the safety, efficacy, and long-term results of this technique.

## **Materials and Methods**

We conducted a retrospective analysis of 11 patients undergoing endovascular stent graft placement for UAF at our institution from 2004 to 2012. Mean age was  $72.8 \pm 11.6$  (range 52–88) years. Female patients were predominant (7 women, 64 %). Patient characteristics, comorbidities, and clinical presentation were reviewed from medical records. In all cases, the fistula was assessed by contrast-enhanced multidetector computed tomography (CECT) to consider the endovascular treatment, and threedimensional reconstruction images obtained by dynamic multiphase scans were evaluated. The clinical diagnosis of UAF and compromised side determination was based on pulsatile ureteral bleeding on cystoscopy and/or massive urinary tract hemorrhage or irregularity of artery on CECT.

All patients initially underwent aortic and pelvic angiography with a 4F pigtail catheter (Medikit, Tokyo, Japan) via the common femoral artery of the uncompromised side. Except for one case of ureteroaortic fistula, a 4F J-shaped catheter (RC-9, Medikit) was inserted into the common iliac artery (CIA) of the compromised side, and multidimensional angiographies were performed. The fistula site was defined as an irregularity of the artery detected on CECT or angiography, including extravasation into the ureter, pseudoaneurysm, and encasement of artery. In cases without arterial irregularity, the fistula site was considered as the crossing area between the ureter and the artery on CECT.

In cases in which the fistula site was located in the aorta, CIA, or external iliac artery (EIA) with an adequate landing zone ( $\geq$  1.5 cm in length), the stent graft was placed in the aorta, the CIA, or the EIA. If the fistula site was located in the CIA without an adequate distal landing zone or in the EIA without an adequate proximal landing zone, the stent graft was placed from the CIA to EIA after coil embolization of the proximal portion of internal iliac artery (IIA) (Fig. 1). If the fistula site was located in the CIA without an adequate proximal landing zone, a bifurcated aortoiliac stent graft was placed.

Details of the hospital course were reviewed. The freedom from recurrent hematuria and the overall and UAFrelated mortality rates were analyzed by the Kaplan–Meier method with SPSS statistical software (version 18.0; IBM, Armonk, NY, USA, USA).

## Results

#### **Clinical Findings**

Clinical findings are summarized in Table 1. Gross hematuria was present in all patients, and previous intermittent mild hematuria had been present in 6 cases (55 %). Chronic ureteral stenting was present in 10 cases (91 %), pelvic surgery in 8 (73 %), pelvic radiation in 5 (45 %), and iliac artery aneurysm in 1 (9 %). The mean period of ureteral stenting before UAF presentation was  $5.2 \pm 4.8$ (range 1–15) years. In 9 cases (92 %), blood transfusion was required. Among these, patients were in hypovolemic shock but blood pressure was maintained by blood transfusion in 5 cases (45 %), and 1 patient (9 %) remained in hypovolemic shock despite the blood transfusion until the procedure.



Fig. 1 A, B CECT revealed left urinary tract hemorrhage (arrowhead; the patient had duplication of the urinary tract). CECT and angiography revealed irregularity of the left CIA (arrow). C Stent

graft placement (Niti-S ComVi,  $10 \times 60$  mm) was performed from the CIA to the EIA after coil embolization for the proximal portion of the IIA

Table 1	Clinical	findings

Patient no.	Age/ sex	Underlying disease	Pelvic surgery	Pelvic radiation	Ureteral stenting (duration, years)	Gross hematuria	Previous mild hematuria
1	76/F	Cervix cancer	Total hysterectomy	Yes	Yes (4)	Yes	Yes
2	86/M	Retroperitoneal fibrosis	_	No	Yes (15)	Yes	Yes
3	88/F	Cervix cancer	Total hysterectomy	Yes	Yes (7)	Yes	Yes
4	59/F	Cervix cancer	_	No	Yes (5)	Yes	Yes
5	82/M	Bladder cancer	Total cystectomy, ureterocutaneous fistula formation	No	Yes (3)	Yes	Yes
6	83/F	Common iliac artery aneurysm	-	No	No	Yes	No
7	70/M	Bladder cancer	Total cystectomy, ureterocutaneous fistula formation	No	Yes (2)	Yes	Yes
8	71/F	Rectal cancer	Lower anterior resection	No	Yes (1)	Yes	No
9	62/F	Retroperitoneal liposarcoma	Incomplete resection	Yes	Yes (2)	Yes	No
10	72/M	Rectal cancer	Total pelvic exenteration, ileal conduit urinary diversion	No	Yes (1)	Yes	No
11	52/F	Cervix cancer	Total hysterectomy	Yes	Yes (12)	Yes	No
Total	-	_	8 (72 %)	5 (45 %)	10 (91 %)	11 (100 %)	6 (55 %)

# **Imaging Findings**

Imaging findings and procedures are summarized in Table 2. Arterial irregularity was revealed in 6 cases (55 %), 3 (27 %) by CECT and 5 (45 %) by angiography (among these, in 2 cases, both imagings were diagnostic). In 5 patients (45 %) with no arterial irregularity, the fistula site was considered to be at the crossing area between ureter and artery on CECT. In the case of a CIA aneurysm, the fistula site was considered to be at the aneurysm. In total, the fistula was located at the EIA in 6 cases (55 %), the CIA in 4 (36 %), and the abdominal aorta in 1 (9 %).

## Procedures

Four procedures (36 %) were performed emergently. Nine procedures (82 %) were performed under local anesthesia and 2 (18 %) under general anesthesia. The device was introduced through the common femoral artery by surgical exposure in 5 (45 %) and percutaneously in 6 (55 %).

A tubular stent graft was placed from the CIA to EIA in 6 cases (55 %), the EIA in 2 (18 %), the CIA in 1 (9 %), and the abdominal aorta in 1 (9 %). A Y stent graft was used in 1 case of CIA aneurysm (9 %). In 8 patients (73 %), the ipsilateral proximal portion of IIA was

Table 2 Imaging findings and procedures

	Site of fistula			SG placement						
Patient no.	CECT	Angiography	Crossing area between ureter and artery	Type of SG	SG length (mm)	SG diameter (mm)	PLZ length (mm)	DLZ length (mm)	Covered area	Embolization of IIA
1	Unknown	EIA	CIA-EIA	Niti-S	40	10	20	20	EIA	Yes
2	Unknown	Unknown	CIA	Zenith	93	12–10 <sup>a</sup>	30	63	CIA– EIA	Yes
3	EIA	Unknown	EIA	Zenith	93	12	58	35	CIA– EIA	Yes
4	Unknown	Unknown	EIA	Niti-S	100 (70.40) <sup>b</sup>	10	48	42	CIA– EIA	Yes
5	Unknown	Aorta	Aorta	Z	50	26	35	15	Aorta	_
6	Unknown	Unknown	CIA	Excluder	160	10	92	30	Aorta– EIA	Yes
7	CIA	CIA	CIA	Niti-S	60	10	20	20	CIA– EIA	Yes
8	Unknown	EIA	EIA	Niti-S	70	8	35	35	EIA	_
9	Unknown	Unknown	EIA	Fluency	56	10	28	28	CIA– EIA	Yes
10	Unknown	Unknown	CIA	Niti-S	40	8	20	20	CIA	_
11	EIA	EIA	EIA	Excluder	140 (100.70) <sup>b</sup>	10	90	40	CIA– EIA	Yes
Total/ mean <sup>c</sup>					84.2	11.2	43.3	31.6		8 (73 %)

SG stent graft, CECT Contrast-enhanced computed tomography, PLZ proximal landing zone, DLZ distal landing zone, CIA common iliac artery, EIA external iliac artery, IIA internal iliac artery, Niti-S Niti-S ComVi, Zenith Zenith iliac limb, Z custom-made stent graft composed of a Z stent and a Dacron graft, Excluder Gore Excluder endograft

<sup>a</sup> Zenith iliac limb was tapered; the proximal and distal diameters were 12 and 10 mm, respectively

<sup>b</sup> Two stent grafts were placed

<sup>c</sup> Six (55 %) were detectable

embolized with 0.018-inch coils (Tornado; Cook, Bloomington, IN) and/or interlocking detachable coils (Boston Scientific Japan, Tokyo, Japan) before the stent graft placement. The mean diameter and length of stent grafts were 11.2  $\pm$  5.0 (range 8–26) mm and 84.2  $\pm$  38.4 (range 40–160) mm, respectively. The mean lengths of the proximal and distal landing zones were 43.3  $\pm$  26.4 (range 20–92) mm and 31.6  $\pm$  13.7 (range 15–63) mm, respectively.

Different types of stent graft were used: Niti-S ComVi (Taewoong Medical, Seoul, Korea) in 5 (45 %), Zenith iliac limb (William Cook Europe Aps, Bjaeverskov, Denmark) in 2 (18 %), Gore Excluder endograft (Gore, Flagstaff, AZ) in 2 (18 %), Fluency (Bard Peripheral Vascular, Tempe, AZ) in 1 (9 %), and a custom-made stent graft composed of a Z stent (William Cook) and a Dacron graft (Ube Kosan, Yamaguchi, Japan) in 1 (9 %). The Niti-S ComVi covered stent, which is a commercial device but available only as a biliary stent, had mainly been used until commercial devices became available in Japan.

#### Early Outcomes

All procedures were successful, and hematuria disappeared in all cases. Early and long-term outcomes are summarized in Table 3. There were no stent graft–related complications. One patient died of urosepsis induced by exchange of ureteral catheter 37 days postoperatively.

# Long-term Outcomes

During the mean follow-up period of 548 (range 35–1,386) days, there were no stent graft–related complications such as thrombosis, kinking, and migration. All stent grafts were patent. Gross hematuria was not observed. However, 4 patients (36 %) had recurrent mild hematuria. The freedom from hematuria recurrence rate at 1 and 2 years was 76.2 and 40.6 %, respectively (Fig. 2).

In patient 1, angiography precisely demonstrated the bleeding site, so the stent graft was placed only in the EIA. However, hematuria recurred 661 days later, and another

Table 3 Outcomes

Patient no.	Acute complication	Recurrence of hematuria (days)	Second treatment	Survival (days)	Dead or alive	Cause of death
1	_	Yes (661)	SG placement	1,386	Alive	
2	-	-		311	Alice	
3	-	Yes (181)	Supportive care	337	Dead	Retroperitoneal abscess
4	-	-		790	Alive	
5	Urosepsis	_		37	Dead	Urosepsis after changing ureter catheter
6	-	Yes (48)	Surgical conversion	340	Alive	
7	-	-		145	Dead	Renal dysfunction
8	-	-		1,049	Alive	
9	-	Yes (623)	Supportive care	623	Dead	Entero-arterial fistula
10	-	-		970	Dead	Multiple lung metastasis
11	_	-		35	Alive	
Total/ mean	1 (9 %)	4 (36 %)		450		

SG stent graft

stent graft was placed from the CIA to the EIA, including the previously treated area. In patient 6, who had a huge saccular CIA aneurysm treated with Y stent graft, hematuria recurred 48 days afterward and was treated with surgical reconstruction, but the fistula site could not be identified in the open field. There was no recurrent hematuria thereafter in both patients. The other 2 patients with recurrence (patients 3 and 9) received supportive care until death. No additional procedures were performed because the hematuria was mild, neither had a recurrent fistula observed on CECT, and general condition and prognosis were poor.

Five patients died during follow-up. One patient died of UAF-related cause. The retroperitoneal abscess may have been induced by the repeated urinary tract or nephrostomy



Fig. 2 Rate of freedom from recurrence of hematuria

tube obstruction caused by clots during recurrent hematuria. Another 4 patients died of non-UAF-related causes: urosepsis (as described above), enteroarterial fistula, chronic renal failure, and multiple lung metastases. The rates of freedom from UAF-related and overall mortality were 85.7 and 68.6 % at 1 year, and 85.7 and 54.9 % at 2 years, respectively (Fig. 3).

## Discussion

The incidence of UAF has been increasing in the past few decades. Before 1994, there were only 33 cases reported, but there are now over 100 cases reported [1]. Possible factors include a better-developed system of registration,



Fig. 3 Freedom from UAF-related and overall mortality rate

improved clinical awareness, and higher incidence of UAF risk factors combined with longer survival [3, 6, 7]. Risk factors for UAF include extensive pelvic surgery (68–100 %), pelvic radiation (46–74 %), chronic ureteral stenting (65–84 %), and peripheral vascular disease (19–42 %). Among these factors, the former three appear to be the most important [1, 5, 6]. The fibrotic changes induced by the inflammatory response to pelvic surgery or radiotherapy cause adhesion between the ureter and the artery. The chronic compression from ureteral stents with arterial pulsation weakens the walls of both the artery and the ureter and finally forms a fistula [10, 13].

Ureteroarterial fistula patients usually present with gross hematuria as the most common symptom, often preceded by an intermittent mild hematuria, which was the case in our study as well [3, 4, 6, 7, 12, 15–17, 19]. In patients with history of radiation, pelvic surgery, and ureteral stenting, it is necessary to suspect UAF, even if the intermittent hematuria is mild. CECT and angiography are first-line diagnostic modalities. However, the reported diagnostic rates of these imaging modalities have been only 42-50 and 63-69 %, respectively [4, 7], which is similar to our experience. A possible reason is that contrast extravasation into the ureter will not occur when a ureteral stent or clots are at the site and tamponade the leak. On the other hand, provocative angiography by removing the ureteral stent over a guide wire or a to-and-fro movement of the ureteral balloon catheter, as well as selective catheterization of the fistula from the artery were reported to produce a diagnostic rate as high as 63-100 % [4, 6, 20]; however, it is potentially dangerous and should be performed with careful preparation.

Recently, endovascular stent graft placement has been recognized as an effective and less invasive treatment of UAF, allowing an early recovery. This treatment also offers advantages over open surgical repair or coil embolization of the iliac artery with extra-anatomical bypass, including closure of the fistula while maintaining the anterograde iliac flow without deterioration of the compromised ureter or adjacent structures [13]. Fox et al. [6] reported that the endovascular success rate was 85 % in 14 cases, and van den Bergh et al. [4] reported no deaths after endovascular treatment in the systematic review of 32 cases. Furthermore, multiple reports of endovascular repair demonstrated short-term good results [9, 11-18]. Our experiences also demonstrate that endovascular stent graft placement can be successfully performed and can control the hemorrhage.

There are no extensive reports regarding the long-term results, but the recurrence rate of hematuria is suspiciously high. Fox et al. [6] reported hematuria recurrence in 2 (14 %) of 14 patients treated with endovascular procedures. Their recurrence rate is rather low; however, the

median follow-up was as short as 4.5 months. In our study, however, the recurrence rate was similar during the first year, but increased during the second year. The ongoing process of inflammation or advancement of malignancy could cause ureteroarterial adhesion or another fistula in the chronic phase. Placing the stent graft to cover a segment longer than the compromised side could be one way to prevent the recurrence. In all of our cases and in some previously reported cases of recurrence, hematuria was mild and did not extend to life-threatening hemorrhage. Thus, close and long follow-up after the procedure is important.

Previous study reported overall and UAF-related mortality rates of 47 and 10 % for open surgery or endovascular treatment, respectively, with a median follow-up of 13 months [6]. If the general condition of the patient allows, additional interventional treatment can be performed and related death prevented, such as death due to recurrent hemorrhage leading to further complications, as seen in our case. It seems unavoidable that the overall mortality rate is high as a result of old age, malignancy, and complications of the underlying disease, but endovascular stent graft treatment could reduce UAF-related death.

In conclusion, endovascular stent graft placement for UAF was a safe and effective method to manage acute events and prevent cause-related death, although mild hematuria frequently recurred during follow-up. Close and long observation after the endovascular procedure is important to timely perform additional treatment.

**Conflict of interest** The authors declare that they have no conflict of interest.

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