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CASE REPORT

Aortoiliac aneurysm with congenital right pelvic kidney

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Abstract The association of congenital pelvic kidney with abdominal aortoiliac aneurysm is an extremely rare clinical finding. Previous reports have described various methods of aneurysm repair with successful preservation of the function of pelvic kidney. However, to our knowledge, reconstruction of more than two renal arteries has not been established. We report a case of abdominal aortic aneurysm complicated by congenital right pelvic kidney in a 72-year-old man. Computed tomography (CT) revealed an abdominal aortic aneurysm with a maximum diameter of 54 mm and a right common iliac aneurysm of 45 mm. In addition, he had a congenital right pelvic kidney and CT angiography identified three right pelvic renal arteries. The upper artery originated from the bifurcation of the terminal aorta and the lower two originated from the right common iliac artery. Three-dimensional CT was helpful for the accurate planning of the operation. Open surgical repair of the aortoiliac aneurysm with a Dacron bifurcated graft replacement was decided and reimplantation of all three right pelvic kidney arteries to the right limb of the graft was also performed. For renal preservation, the right pelvic kidney arteries were perfused with cold Ringer's lactate using a rapid infusion pump and coronary perfusion cannula. The patient's postoperative course was uneventful, and worsening of renal function was not observed. The perfusion of renal arteries with cold Ringer's solution was thought to be a simple and appropriate procedure for renal protection.

Keywords Abdominal aortic aneurysm · Congenital pelvic kidney · Renal ischemia

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Introduction

The incidence of pelvic kidneys is 1 out of 2100–3000 births in the population [1]. The complication of pelvic kidney with aortoiliac aneurysm is a rare clinical finding, but repair of the aneurysm is challenging because supra-renal aortic clamping is required in most cases. Previous reports have demonstrated various methods of repair without worsening of pelvic kidney function. Only three cases of open repair have been reported in Japan, and a total of 28 cases have been published worldwide. None of the reports described the simultaneous reconstruction of more than two pelvic kidney arteries. We present a case of successful abdominal aortic aneurysm (AAA) repair with revascularization of a right pelvic kidney. In addition, we review 28 other cases from previous reports of this rare pathologic association.

Case report

A 72-year-old man with common atrial flutter was referred to our institution for the treatment of an asymptomatic AAA and right common iliac artery (CIA) aneurysm. He received flecainide, digoxin, and bisoprolol as anti-arrhythmic therapy and warfarin as anticoagulation therapy. He complained of frequent palpitations during his daily activities, so he requested further treatment for the

arrhythmia. However, it was impossible to perform catheter ablation therapy because of the aneurysm. Besides, the size of both the aneurysms was indicative of treatment because of a high risk of rupture. A computed tomography (CT) scanning revealed an aortoiliac aneurysm and a right pelvic kidney. The maximum diameter of the AAA was 54 mm and that of the right CIA aneurysm was 45 mm. In addition, CT angiography revealed that the upper right pelvic kidney artery arose from the bifurcation of the terminal aorta and the two lower arteries arose from the right CIA aneurysm (Fig. 1). Preoperative serum blood urea nitrogen and creatinine levels were 25.5 and 1.13 mg/dL, respectively. Cardiac catheterization revealed no stenosis in the coronary arteries, and transthoracic echocardiography showed good cardiac function. After strict analyses, the patient's cardiac function was confirmed apt to withstand surgery. Although endovascular repair was considered, our institution does not use branched or fenestrated endovascular grafts. Thus, we decided to repair the aneurysm by open surgery and use selective cold perfusion for pelvic kidney protection during aortic clamping. In addition, we planned to revascularize all pelvic kidney arteries if possible. As for anticoagulation therapy, warfarin was suspended 4 days before the surgery, and intravenous unfractionated heparin treatment was initiated from that day onward.

After induction of general anesthesia, we performed midline laparotomy and transperitoneal exposure of the aorta. The infrarenal aorta, both right and left common iliac arteries, the inferior mesenteric artery and all three of the right pelvic arteries were isolated. Following systemic heparinization, proximal aortic cross-clamping was performed distal to the left renal artery and distal clamping was performed just proximal to the bifurcation of the external and internal iliac arteries on both sides. The aneurysm was incised longitudinally and the thrombus was removed. We identified the orifices of the three pelvic kidney arteries. We cannulated the arteries with a coronary perfusion cannula (Coronary Perfusion Cannulae. Self-inflating balloon tip, right angle, MAQUET Japan K.K., Tokyo, Japan). During the cross-clamping, a total bolus infusion of 500 mL of cold Ringer's lactate (Lactec Injection®: Otsuka Pharmaceutical Factory, Inc. Tokushima, Japan) was administered through the cannulas every half hour by a rapid infusion pump. The aneurysm was replaced by an 18 × 9 mm Dacron bifurcated graft (Triplex®: TERUMO corporation, Tokyo, Japan). The selective cold perfusion of the pelvic kidney arteries was continued intermittently until their separate reimplantation to the right iliac limb of the graft after the proximal aortic anastomosis and the right distal iliac anastomosis. The left iliac anastomosis was performed under direct hematic perfusion of the pelvic kidney arteries. Finally, we reimplanted the inferior mesenteric artery to the main body of the graft by side-clamping the graft because the diameter of the artery was about 3 mm.

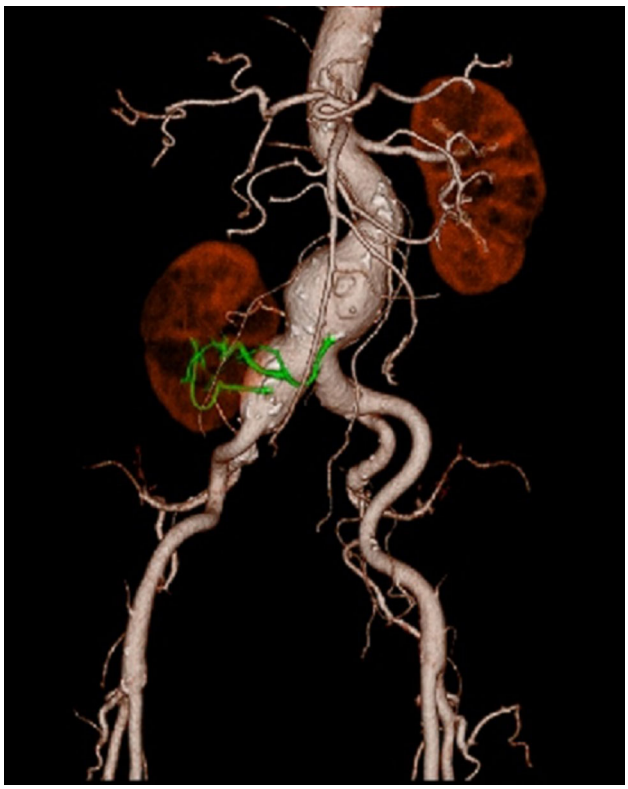

Fig. 1 Reconstructed three-dimensional computed tomography image showing the preoperative aneurysm. Three pelvic kidney arteries are visualized. The upper one originated from the terminal aorta and the two lower ones from the right common iliac aneurysm

The total operation time was 287 min. The ischemic time of the pelvic kidney arteries because of pelvic kidney artery anastomoses was 22 min. We used autologous blood transfusion by cell salvage during the surgery and the bleeding amounted to approximately 300 mL. Hence, allogeneic blood transfusion was not needed. The patient was extubated in the operation room after the procedure was completed, and the postoperative course was uneventful. Anticoagulation therapy with heparin infusion was initiated on postoperative day (POD) 1 and with oral warfarin on POD 2 after oral intake was restarted. Heparin infusion was continued until the targeted warfarin control was reached. CT scanning was performed on POD 8; all the pelvic kidney arteries reimplanted to the graft were patent and renal infarction was not observed (Fig. 2). The patient presented with hypouresis several days after the surgery probably because of transient renal function deterioration, which improved after treatment with a diuretic drug (furosemide). Thus, the patient's condition and renal function did not worsen. Serum blood urea nitrogen and creatinine levels were 14.2 and 1.02 mg/dL, respectively,

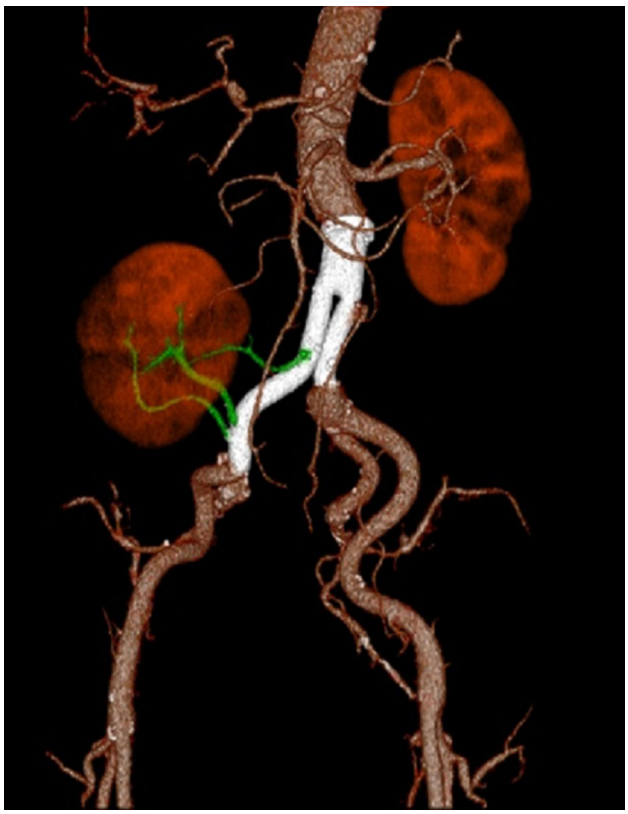

Fig. 2 Postoperative three-dimensional reconstruction computed tomography. Dacron bifurcated graft is presented in *white*. All pelvic kidney artery reimplantations to the right iliac limb of the graft are patent. Renal infarction is not revealed

on the day of hospital discharge. At the 3-month-followup examination at the outpatient clinic, no deterioration was noted in the patient's renal function.

Discussion

Recently, the diagnosis and treatment of aortic disease has progressed dramatically due to the development of diagnostic technique and therapies. Nakayama et al. reported that the 30-days operative mortality after elective AAA repair was 1.2 % (10/869). No significant preoperative predictor of postoperative mortality could be identified within 30 days after surgery [2]. Weakening of the aortic wall is deeply involved in the pathogenesis of aortic aneurysm. However, risk factors of AAA other than hypertension remain to be determined. Saruhara et al. [3] showed that the incidence of moderate to severe obstructive sleep apnea was significantly higher in the AAA patients.

Various congenital renal anomalies have been reported, such as anomalies of migration (pelvic, lumbar, abdominal, iliac and thoracic) [4] and anomalies of fusion (horseshoe [5] and crossed [6]). The most common variant is an anomalous vascular supply to the pelvic kidney. One or two renal pelvic kidney arteries are common and usually arise from the aortic bifurcation, the CIA, and the external iliac artery. Rarely, blood supply is branched from the inferior mesenteric artery. These anatomical anomalies are ordinarily asymptomatic [7]. The combination of congenital pelvic kidney with an aortoiliac aneurysm is extremely rare; a radiological study reported that 0.18 % of patients who underwent a major aortic surgery had pelvic kidneys [8]. In 1977, Ezzet et al. [9] presented the first case of a patient with a pelvic kidney associated with AAA. Since then, according to the results of our literature search, a total of 28 cases of aortic aneurysm repair in association with congenital pelvic kidney have been reported (Table 1) [7–28]. Among these reports, three cases involved endovascular repair [7, 15, 25]. However, to the best of our knowledge, reports on reconstruction of more than three pelvic kidney arteries have not been published yet. Hence, we believe that this case is unique.

Conventional arterial angiography is a more invasive procedure, but recently, CT has progressed to provide accurate information about both aortoiliac aneurysms and pelvic kidney arteries. CT angiography and three-dimensional CT can identify even small branches or accessory renal arteries that may go unnoticed via other diagnostic imaging techniques. Regardless of the method used for aneurysm repair, whether it is open surgery or endovascular repair, preoperative planning for revascularization of the pelvic kidney and renal protection is thought to be important. Renal function after aneurysm repair is generally affected by intraoperative renal ischemia. Postoperative renal infarction may also deteriorate renal function and cause acute pain or fever. The renal artery diameter is a key aspect in the decision making concerning revascularization of pelvic kidney arteries. Renal arteries < 3 mm in diameter can usually be sacrificed without any postoperative complications if the preoperative renal function is within normal limits and the main renal artery functions efficiently [25]. In the present case, the upper pelvic kidney artery branched out from the bifurcation of the terminal aorta and the lower two arteries originated from the right CIA aneurysm. The lowest artery had a smaller diameter than the two upper ones, but its perfusion area seemed to be broad. Still, we were able to cannulate all arteries with a coronary perfusion cannula. Thus, we decided to revascularize all three pelvic kidney arteries separately to lower the risk of renal infarction. A postoperative diuretic was administered for maintenance of adequate urinary output to prevent worsening of renal function.

It is essential to prevent intraoperative renal ischemia to preserve renal function. Several procedures mainly for use during aneurysmectomy in renal transplant patients

Table 1 Literature review of cases of aortoiliac aneurysm repair performed in the presence of congenital pelvic kidney

References	No. of patients	Side	Type of aortic repair	Renal protection technique
Ezzet et al. [9]	1	Left	Dacron bifurcated graft	Simple clamping
Hans et al. [10]	1	Right	Dacron bifurcated graft	Selective cold perfusion
Hollis et al. [11]	2	1 Right	Dacron bifurcated graft, 2 of 2 pelvic kidney arteries reimplanted to the main body of the graft	Proximal double clamping during proximal anastomosis and in situ cold perfusion during distal anastomosis
		1 Left	Dacron tube graft, the lower of 2 pelvic kidney arteries included in the distal aortic anastomosis and the upper reimplanted in the left common iliac artery	Selective cold perfusion
Belcastro et al. [12]	1	Right	Dacron tube graft	Distal double clamping
Shchneider et al. [13]	1	Left	Dacron bifurcated graft, 1 of 1 pelvic kidney artery reimplanted to the right iliac limb of the graft	Proximal double clamping during proximal anastomosis and temporary shunt from the body of the graft to the pelvic kidney artery during distal iliac anastomoses
Glock et al. [14]	1	Right	Dacron tube graft, 1 of 2 pelvic kidney arteries reimplanted, the other was included in the distal anastomosis.	Distal double clamping
Kaplan et al. [15]	1	Solitary	Endovascular device	–
Rehrig et al. [16]	1	Right	Dacron bifurcated graft, 1 of 1 pelvic kidney artery reimplanted	Proximal double clamping during proximal anastomosis and temporary shunt from the main body into the pelvic kidney artery during iliac anastomoses
Faggioli et al. [8]	3	NA	Ectopic renal arteries reimplanted or included in the distal anastomosis	Selective cold perfusion if cross-clamp time > 40 min, and no protection if the time < 40 min
Renzulli et al. [17]	1	Horse shoe	Dacron tube graft with superior mesenteric artery selective bypass graft	Proximal double clamping during proximal anastomosis and selective cold perfusion to the largest renal artery during distal anastomosis
Murakami et al. [18]	1	Solitary	Dacron tube graft, 2 pelvic kidney arteries included in the distal anastomosis	Selective cold perfusion to the upper pelvic kidney artery
Hanif et al. [19]	1	Right	Dacron trifurcated graft, selective grafting of 1 of 1 pelvic kidney artery (9 mm graft)	Temporary shunt from the right axillary artery to the pelvic kidney artery during all anastomoses
Mandolfino et al. [20]	1	Right	Dacron bifurcated graft, 2 of 2 pelvic kidney arteries reimplanted to the graft	Simple clamping with systemic administration of dopamine and mannitol
Bui et al. [28]	1	Left	Dacron bifurcated graft, 1 of 1 pelvic kidney artery reimplanted to the graft.	Simple clamping with cooling
	1	Left	Dacron tube graft	Simple clamping
Coney et al. [21]	1	Left	Dacron bifurcated graft, 2 of 2 pelvic kidney arteries reimplanted to the right iliac limb of the graft.	Temporary shunt from the right axillary artery to the pelvic kidney arteries during proximal and right iliac anastomosis
Marone et al. [7]	4	Right	Dacron tube graft, 1 of 1 pelvic kidney artery included distal anastomosis	Selective cold perfusion
		Left	Dacron tube graft, 1 of 1 pelvic kidney artery reimplanted to the graft	Selective cold perfusion
		Right	Dacron bifurcated graft, 1 of 1 pelvic kidney artery reimplanted to the main body of the graft.	Selective cold perfusion
		Left	Dacron bifurcated graft, 1 of 2 pelvic kidney arteries reimplanted to the main body of the graft, the other was included in the distal anastomosis.	Selective cold perfusion

Table 1 continued

References	No. of patients	Side	Type of aortic repair	Renal protection technique
Morales et al. [22]	1	Left	Endovascular device	–
Makris et al. [24]	1	Solitary	Dacron tube graft, 1 of 2 pelvic kidney arteries included in the distal anastomosis	Temporary shunt from the right axillary artery to the right femoral artery and a second shunt from the right common iliac artery to the pelvic kidney artery during all anastomosis
Spear et al. [25]	1	Left	Endovascular device	–
Jinnouchi et al. [26]	1	Right	Dacron trifurcated graft, selective grafting of 1 of 1 pelvic kidney artery (8 mm graft)	Selective cold perfusion
Akashi et al. [27]	1	Right	Dacron tube graft, 1 of 2 pelvic kidney arteries reimplanted to the graft	Selective cold perfusion
Present case. (2014)	1	Right	Dacron bifurcated graft, 3 of 3 pelvic kidney arteries reimplanted to the right iliac limb of the graft	Selective cold perfusion
Total	29			

have been reported. These renal protection methods were generally divided into four groups. First, simple clamping and systemic administration of catecholamines (dopamine) and diuretics (furosemide) were performed [20]. These pharmacological agents are thought to induce diuresis before aortic cross-clamping, thereby reducing the risk of acute tubular necrosis. This method may be the simplest and safest procedure if the aortic clamping time is short. Second, a temporary aorto-femoral or axillo-femoral shunt, named Gott shunt [29] may be effective [13, 16, 21, 24]. This method can preserve a physiological, continuous renal perfusion. However, it requires a groin incision, is more invasive, and is associated with complications such as embolization, hemorrhage, dissection and groin complications. In addition, potential complications tend to introduce the risk of graft infection. Third, the double proximal clamping technique, reported in 1986 by Lacombe et al. [30], enables the preservation of renal function during a medium clamping time (may be < 60 min) [11, 12, 14, 16]. With this method, retrograde flow from the lumbar, inferior mesenteric and iliac arteries is spared during proximal anastomosis. However, this technique is limited if the proximal neck of the aneurysm is not long enough to clamp at two sites or if adequate collateral circulation is not established. Fourth, selective cold perfusion was used in the majority of cases [7, 8, 10, 11, 17, 18, 26, 27]. Renal preservation by cold Ringer's lactate has become more frequent in the field of renal transplantation, and this technique is often employed for suprarenal clamping when longer clamping times (approximately 60–90 min) are needed. Two approaches were reported; one consisted of a bolus infusion every 20–30 min and the other consisted of a continuous infusion. Which of the two approaches has better results is left to the clinician's consideration. Both approaches are simple but may have limitations in terms of renal ischemic time. Another way to achieve renal perfusion is through extracorporeal circulation [31]. It allows longer renal ischemic time, but this method requires a perfusionist and a pump oxygenator. A groin incision for retrograde cannulation into the femoral artery and femoral vein is forcefully required as well.

In our department, when suprarenal clamping is required during aortic surgery, the approach employed is selective cold perfusion with bolus infusions of Ringer's lactate solution every 30 min. In the present case, we used the same procedure for all pelvic kidney arteries. In addition, the pelvic kidney arteries had a smaller diameter than a normal renal artery, so we used coronary perfusion cannulas that were just the right size for cannulation.

In recent years, developments in endovascular repair of pararenal, juxtarenal, and thoracoabdominal aneurysm have been remarkable. As previously noted, only three cases of aortic aneurysm repair with congenital pelvic kidney have been reported to date. Yet, endovascular repair for such cases will presumably increase in the next decades. Branched or fenestrated devices provide a safe option compared with open surgery. In our institution, we lack experience in the use of such devices. Therefore, our choices were limited and we decided to perform open surgery. In the future, as the endovascular technique continues to advance and grow more common, patients could be offered this additional therapeutic alternative.

Conclusion

Aortoiliac aneurysm repair with congenital pelvic kidney requires strict preoperative planning with the use of accurate imaging studies, e.g., three-dimensional CT. To protect renal function in the peri- and postoperative period, various methods have been reported. We found that selective cold perfusion was used in most cases. To date, a therapeutic gold standard technique has not been established. The most adequate method should be chosen in a case-by-case basis considering each patient's condition and the operative approach implemented.

In our case, we revascularized all three pelvic kidney arteries and used selective cold perfusion during the aortic repair. As a result, renal function did not deteriorate, and our approach toward this case was appropriate and effective.

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