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Case Report

Three cases of recurrences after stent-graft placement for arterio-visceral/arterio-luminal fistulas in long-term follow-up [☆]

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ABSTRACT

We report 3 patients with recurrences after stent-graft placement for arterio-visceral/arterio-luminal fistulas in long-term follow-up. Two patients had ureteroarterial fistulas and the other had a tracheo-innominate artery fistula. All 3 patients had hemorrhage on initial presentation and underwent a stent-graft placement for an arterio-visceral/arterio-luminal fistula. Recurrences occurred over a period of 8–26 months and were diagnosed by contrast-enhanced computed tomography; pseudoaneurysms were found in contrast-enhanced computed tomography images in all cases. Pseudoaneurysms may be noted on contrast-enhanced computed tomography as the only finding of recurrences during long-term follow-up after stent-graft placement for arterio-visceral/arterio-luminal fistulas.

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Introduction

Ureteroarterial (UAF), tracheo-innominate artery (TIF), and aortoenteric fistulas are rare conditions in which an artery contacts the surrounding luminal organs to form a fistula. They show similarities in their pathogenesis, predisposing factors, and clinical symptoms and are, therefore, often collectively known as arterio-visceral/arterio-luminal fistulas [1]. Recently, endovascular treatment for arterio-visceral/arterio-luminal fistulas, mainly by stent-graft placement, has increased [2,3]. However, long-term outcomes after stent-graft placements for arterio-visceral/arterio-luminal fistulas are still unclear. Because few reports exist on the recurrence of arterio-visceral/arterio-luminal fistulas [4], computed tomography (CT) findings are still unclear. In this paper, we reported 3 cases of CT findings and clinical data of recurrences after stent-graft placement for arterio-visceral/arterio-luminal fistulas in long-term follow-up.

Case reports

Case 1

A 53-year-old female patient was treated for cervical cancer with heavy particle irradiation, anterior pelvic exenteration, ileal conduit surgery, and a bilateral ureteral stent placement. Gross hematuria was observed during the left ureteral stent placement. Contrast-enhanced CT (CECT) revealed a stenosis of the left common iliac artery crossing with the left ureteral stent, which was diagnosed as UAF. An 8 × 50 mm Viabahn stent-graft (WL Gore and Associates, Flagstaff, AZ, USA) was placed to seal the UAF (Fig. 1A). The landing zone, which is the portion of a blood vessel where the stent-graft is positioned, was 17 mm proximally and 27 mm distally from the fistula. Gross hematuria was no longer observed immediately after the stent-graft placement but was observed 8 months later. CECT revealed a pseudoaneurysm with periarterial soft tissue thickening at the proximal edge side of the stent-graft, which was at the origin of the left common iliac artery (Fig. 1B). The pseudoaneurysm was 10 × 12 mm in size in CT images. Moreover, CECT revealed the destruction of an adjacent vertebral body and deformation of the stent-graft. These findings led to a diagnosis of UAF recurrence. An 8 × 50 mm Viabahn stent-graft was placed from the terminal aorta to the right common iliac artery and an 8 × 100 mm stent-graft was placed from the terminal aorta to the left common iliac artery. Subsequently, two 7 × 40 mm balloons (Rx-Genity, Kaneka Medical Products, Osaka, Japan) were postdilated using a kissing balloon technique. Gross hematuria was no longer observed immediately after the procedure. Although blood cultures were negative, antibiotics were used for life-long therapy due to suspicion of infection. No antiplatelet therapy was performed because of a history of embolization for pelvic hemorrhage. However, the patient subsequently died due to the aggravation of underlying disease 7 months afterwards.

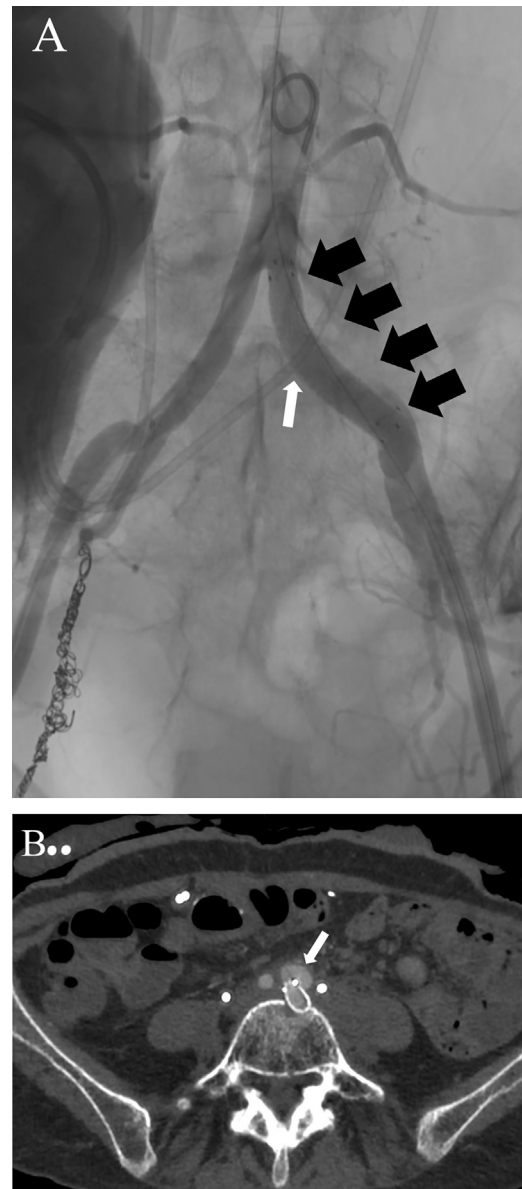


Fig. 1 – (A) An 8 × 50 mm Viabahn stent-graft (black arrows) was placed so that the left common iliac artery could seal the ureteroarterial fistula (white arrow). The patient had a history of embolization of the right inferior gluteal artery due to pelvic bleeding. (B) Contrast-enhanced computed tomography (CECT) image obtained 8 months after the placement of a stent-graft revealed a pseudoaneurysm (white arrow) with periarterial soft tissue thickening at the proximal edge side of the stent-graft, which was at the origin of the left common iliac artery.

Case 2

A 38-year-old female patient was treated for cervical cancer with heavy particle irradiation, a para-aortic lymphadenectomy and a right ureteral stent placement. In addition, the patient had a history of perforation of the sigmoid colon after heavy particle irradiation and para-aortic lymphadenectomy, leading to a permanent stoma. Gross hematuria was observed

during the ureteral stent replacement. CECT revealed the right ureteral stent and right external iliac artery were only in contact, with no imaging findings observed such as a stenosis or pseudoaneurysm. Angiography showed similar findings. However, because we strongly suspected clinical UAF where the right ureteral stent contacted the right external iliac artery, a 6 × 50 mm Viabahn stent-graft was placed to seal the suspected UAF (Fig. 2A). The landing zone length from the fistula was 14 mm proximally and 32 mm distally. Gross hematuria was no longer observed immediately after the stent-graft placement. However, gross hematuria was observed 20 months later. CECT revealed a pseudoaneurysm with periarterial soft tissue thickening at the proximal edge side of the stent-graft, which was at the origin of the right external iliac artery (Fig. 2B). The pseudoaneurysm was 2 × 5 mm in size with an indistinct and irregular appearance. These findings led to a diagnosis of UAF recurrence. Endovascular treatment was selected because the patient was unlikely to endure surgical treatment. Each of 7 × 50 mm and 7 × 25 mm Viabahn stent-grafts were placed from the right common iliac artery to the right external iliac artery after embolization of the right internal iliac artery. Antibiotics were used for 30 days because blood cultures revealed *Pseudomonas aeruginosa*. Moreover, dual antiplatelet therapy was used for 6 months. The patient is on single antiplatelet therapy for life. Symptoms or major complications related to the procedure are not noted during 8 months' follow-up.

Case 3

A 36-year-old female patient was admitted to our hospital because of non-Fukuyama congenital muscular dystrophy. The patient had previously undergone a tracheostomy and was fitted with a tracheal cannula. A minor intratracheal hemorrhage was observed on positional change. Subsequently, a massive intratracheal hemorrhage was observed immediately on removal of the tracheal cannula cuff. CECT revealed the tracheal cannula and brachiocephalic artery were only in contact. Because angiography was performed and revealed a pseudoaneurysm in the area of contact, we diagnosed TIF. Each of 8 × 25 mm and 8 × 29 mm VBX Viabahn stent-grafts (WL Gore and Associates) were placed at the brachiocephalic artery to seal the TIF (Fig. 3A). The landing zone length from the fistula were 20 mm proximally and 12 mm distally. Intratracheal hemorrhage was no longer observed immediately after the procedure but was observed 26 months later. CECT revealed a pseudoaneurysm with periarterial soft tissue thickening and surrounding fat stranding at the distal edge side of the stent-graft, which was at the origin of the right subclavian artery (Fig. 3B). The pseudoaneurysm was 12 × 15 mm in size. These findings led to a diagnosis of TIF recurrence. However, the patient died of a rupture of the pseudoaneurysm before any additional treatment could be given. No blood cultures or antiplatelet therapy were performed.

Discussion

We describe 3 cases of pseudoaneurysms in which CECT revealed the recurrence of arterio-visceral/arterio-luminal fistu-

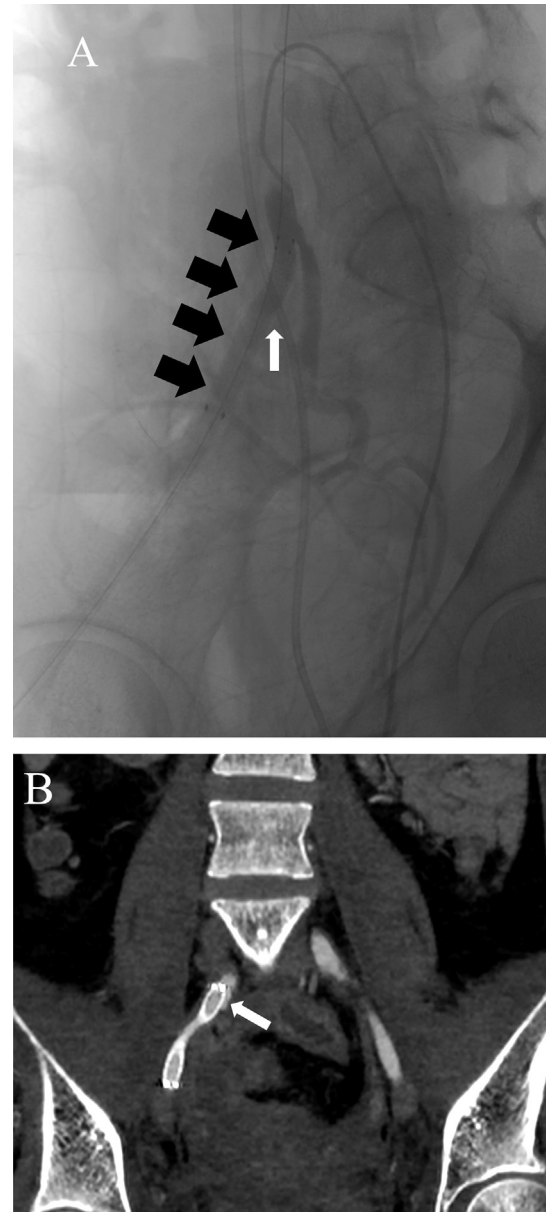


Fig. 2 – (A) A 6 × 50 mm Viabahn stent-graft (black arrows) was placed so that the right external iliac artery could seal the suspected ureteroarterial fistula (white arrow). **(B)** Contrast-enhanced computed tomography (CECT) image obtained 20 months after placement of a stent-graft revealed a pseudoaneurysm (white arrow) with periarterial soft tissue thickening at the proximal edge side of the stent-graft, which was at the origin of the right external iliac artery.

las during long-term follow-up after stent-graft placement for UAF in 2 cases and TIF in 1 case. To date, UAF during long-term follow-up after stent-graft placement have been few reported [2,5–7], and only 1 article has described a pseudoaneurysm with UAF recurrence in the long-term after stent-graft placement occurring in two cases as detected by angiog-

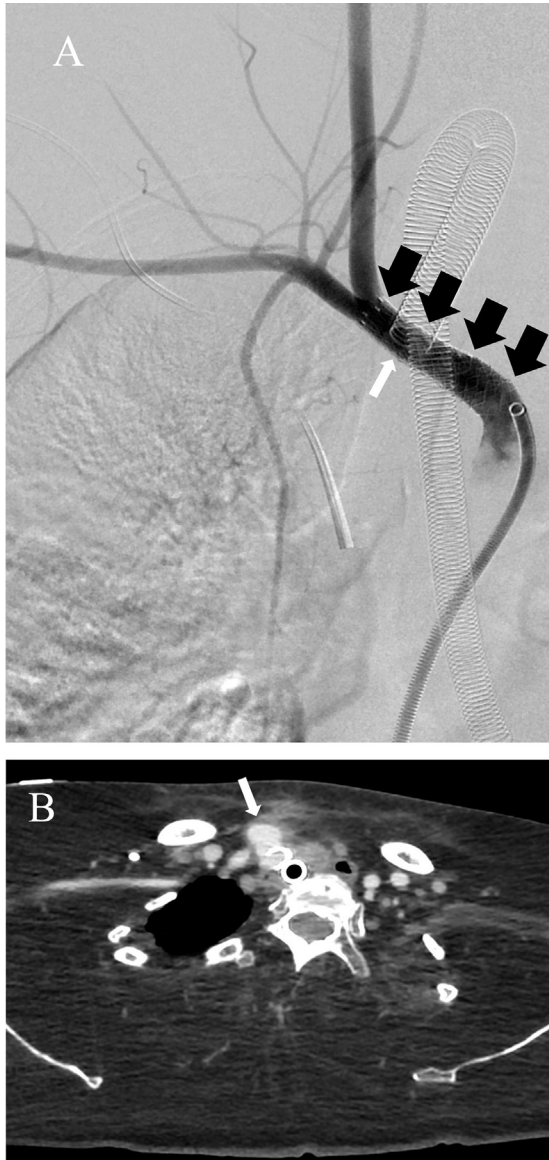


Fig. 3 – (A) 8 × 25 mm (black arrows) and 8 × 29 mm VBX (black arrows) Viabahn stent-grafts were placed at the brachiocephalic artery to seal the tracheo-innominate artery fistula (white arrow). (B) Contrast-enhanced computed tomography (CECT) image obtained 26 months after placement of a stent-graft revealed a pseudoaneurysm (white arrow) with periarterial soft tissue thickening and surrounding fat stranding at the distal edge side of the stent-graft, which was at the origin of the right subclavian artery.

raphy; however, CT findings were not discussed [2]. Although TIF during long-term follow-up after stent-graft placement have been reported as only a case report [8], no reports existed describing a pseudoaneurysm in the long-term after stent-graft placement for TIF. In case 3, although not life-saving, the CECT accurately revealed a pseudoaneurysm. Our report described not only UAF and TIF during long-term follow-up after stent-graft placement, but also CT findings at recurrence with

long-term follow-up after stent-graft placement of arterial-organ/arterial duct fistulas, which were pseudoaneurysms in all 3 cases. As reported, the CT finding at recurrence after stent-graft placement was only a pseudoaneurysm, which suggests this is clinically valuable information, though an initial arterio-visceral/arterio-luminal fistula may present with various CT findings, such as pseudoaneurysms, stenoses or extravasation, or even no CT findings [2,4,9].

Yoshioka et al. described how, when a stent-graft was positioned adjacent to the ureteral stent where infection spread to a normal artery, focal perforation of the arterial wall, and a pseudoaneurysm reoccurred after stent-graft placement for UAF [10]. TIF is similar in pathogenesis and predisposition to UAF with regard to an arterio-visceral/arterio-luminal fistula [1]. We considered the same pathogenesis may be established after stent-graft placement. Furthermore, although blood cultures were positive only in case 2, CT findings in cases 1, and 3 suggested an infection. Therefore, we considered infection to be the main cause of the pseudoaneurysms in our 3 cases.

Ueda et al. reported a Viabahn stent-graft should be secured for at least 20 mm according to the manufacturer's instructions [11]. In addition, they reported that if the diameter of the Viabahn stent-graft was appropriately sized, postoperative type 1 endoleaks did not occur even when the landing zone length could not be secured for 20 mm. However, if infection existed surrounding the target vessels, a higher risk of re-bleeding or endoleaks existed due to the spread of inflammation to the vessel walls [11]. In our cases, the stent-graft was positioned adjacent to the ureteral stent or tracheal cannula because it was challenging to secure the landing zone length for more than 20 mm. In addition, in our cases, pseudoaneurysms were observed at the stent-graft edge side when a landing zone length was less than 20 mm. Therefore, in arterio-visceral/arterio-luminal fistulas, it should be noted that pseudoaneurysms may appear on the edge of a stent-graft with a landing zone length of less than 20 mm on CECT.

Patients may need to be closely followed for up to 1 year after stent-graft placement because of the possibility of recurrence [12]. However, we experienced cases of recurrence more than 1 year after stent-graft placement for arterio-visceral/arterio-luminal fistulas. Therefore, long-term follow-up after stent-graft placement for arterio-visceral/arterio-luminal fistulas may be necessary in addition to close follow-up within the first year after the procedure.

The main limitations of this report were the use of retrospective data and a small sample size. Therefore, a future study with a large number of cases is required to confirm our findings.

In conclusion, pseudoaneurysms may be noted on CECT as the only finding of recurrences after long-term follow-up and stent-graft placement of arterio-visceral/arterio-luminal fistulas.

Ethical approval

This retrospective study was approved by Ethical Board of Kochi University Medical School (decision number of ethics committee approval: 2023-73).

Patient consent

Written informed consent has been obtained from all patients for publication of the case report.

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