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Case Report

External iliac artery pseudoaneurysm following treatment for arterio-ureteral fistula using a balloon-expandable stent-graft: A case report*,**

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ABSTRACT

Sepsis and bleeding can lead to life-threatening complications, such as stent-graft infection and pseudoaneurysm, after stent-graft implantation. An 83 year-old woman was admitted to our hospital for sepsis 14 months after treatment with a balloon-expandable stentgraft for an arterio-ureteral fistula (AUF) between the right external iliac artery and the right ureter. Blood cultures were positive for methicillin-resistant *Staphylococcus aureus* and *Candida tropicalis*. A giant infectious pseudoaneurysm (44×70 mm) at the distal edge of the stent-graft was suspected of having caused the sepsis. Although endovascular therapy (EVT) was planned to correct the pseudoaneurysm, the right iliac artery spontaneously became occluded from the ostium of the right common iliac artery to the common femoral artery 5 days after diagnosing the pseudoaneurysm; hence, EVT was not performed. Antibiotic administration was continued, and blood culture results were negative. Although EVT using a stent graft for AUFs is effective, data on the chronic phase outcomes are limited. Therefore, patients with AUFs treated using stent-grafts should be carefully followed up.

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Introduction

Pseudoaneurysm formation after endovascular therapy using a stent-graft is a relatively rare but potentially life-threatening condition because of the risk of infection and bleeding [1]. Endovascular treatment of arterio-ureteral fistula (AUF) with covered stents has been described [2-3], as endovascular treatment with stents is less invasive and provides rapid control of life-threatening bleeding. However, complications have not been commonly reported after implantation of a stent-graft for treating AUF [4], and data on outcomes in the chronic

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Fig. 1 – CT scan and angiography findings (A) CT findings after stent graft implantation, (B) Angiography findings after stent graft implantation, (C) CT scan showing a huge pseudoaneurysm formation (white arrow) at the right external iliac artery, (D) CT angiography (coronal plane) showing a huge pseudoaneurysm formation (white arrow) at the right external iliac artery, (E) Angiography showing pseudoaneurysm formation (red arrowhead) and floating thrombus (yellow dotted oval), (F) CT scan showing no contrast into the pseudoaneurysm (white arrow), and (G) occlusion of the right iliac artery from the ostium of the right common iliac artery to the right common femoral artery (white arrow), (H) Angiography showing occlusion of the right iliac artery from the ostium of the right iliac artery from the ostium of the right arrow), CT, computed tomography. (Color version of figure is available online.)

phase are limited. Here, we report a rare case of an external iliac artery pseudoaneurysm after treatment for AUF using a balloon-expandable stent-graft.

Case Presentation

An 83 year-old woman underwent balloon-expandable stentgraft implantation for AUF between the right ureter and external iliac artery (EIA) 14 months earlier. Details of this case have been previously reported [5]. The computed tomography (CT) and angiography findings after stent-graft implantation are shown in Figures 1A and 1B, respectively. The patient visited our hospital with complaints of pain and red flares from the right side of the waist to the thigh. She was admitted to the Department of Dermatology on suspicion of cellulitis. Blood culture was performed, and ceftriaxone sodium hydrate (2000 mg/day) was administered. However, the inflammation did not improve, and the blood culture was repeated. The second test was positive for methicillin-resistant Staphylococcus aureus (MRSA). Hence, the antibiotics were changed to vancomycin hydrochloride (750 mg/day) on day 17. On day 40, vancomycin was changed to daptomycin (250 mg/day) because of worsening renal function. On day 45, enhanced CT was performed,

as the inflammation did not improve despite the disappearance of cellulitis. Enhanced CT revealed enhanced peri-stent graft soft tissue and a massive pseudoaneurysm (44×70 mm) in the right EIA (Figs. 1C and 1D). Two days later, angiography was performed to outline the pseudoaneurysm and a possible fracture and/or deformation of the stent-graft. Angiography revealed that the pseudoaneurysm formed at the distal edge of the stent-graft (Fig. 1E). Moreover, we observed a floating thrombus in the stent-graft (Fig. 1E). Although there were no significant stent-graft fractures under fluoroscopy (Fig. 2A, 2B, and 2C), complete deformation of the stent-graft was observed compared to just after its implantation (Fig. 2A and 2D). That same day, a blood culture was performed again, which showed positivity for Candida tropicalis; although MRSA was now negative. Hence, micafungin sodium hydrate (100 mg/day) was included in the therapy. After discussing with the surgeons, we first controlled the bleeding risk of the pseudoaneurysm and subsequently performed EVT, implanting a new stent graft to cover the pseudoaneurysm. However, on day 52, angiography revealed that the right iliac artery had spontaneously become occluded, and blood flow to the pseudoaneurysm was not confirmed (Fig. 1H). Hence, we did not perform any invasive interventions. Additionally, enhanced CT did not show any enhancement of the pseudoaneurysm (Fig. 1F). Contrarily, blood flow to the right lower limb through collateral



Fig. 2 – Detailed view of the stent graft as observed under a fluoroscope (A) Fluoroscope showing stent graft deformation entirely. There are no significant fractures observed in the multidirectional view, (B) Stent graft deformation from the right anterior oblique 40° position, (C) from left anterior oblique 40° position, and (D), just after the stent graft implantation



Fig. 3 – The timeline, including the data of inflammatory reaction and the detail of antibiotics usage. CRP, C-reactive protein ; WBC, white blood cell; CT, computed tomography; CTRX, ceftriaxone sodium hydrate; VCM, vancomycin hydrochloride; DAP, daptomycin; MCFG, micafungin sodium hydrate; SMX/TMP, sulfamethoxazole, trimethoprim; ITCZ, itraconazole

channels from the common femoral artery was confirmed (Fig. 1G).

Discussion

Antibiotics were continued, and the blood culture test result became negative. Antibiotics were changed to oral antibiotics (sulfamethoxazole 1600 mg/day, trimethoprim 320 mg/day, and itraconazole 100 mg/day), and the patient was discharged on day 97. The timeline, including the data of inflammatory reaction and the details of antibiotics usage, is shown in Fig. 3.

Our four discussion points include the reason for the occurrence of the stent-graft infection, the cause of the pseudoaneurysm, the reason behind occlusion of the right iliac artery, and the choice of treatment strategy.

Several reports show that endograft infection occurs in approximately 1% of implantations [6-9]. Lyons et al. reported di-

agnostic criteria for aortic stent graft infection [10]. Based on the criteria, we suspected stent-graft infection in this case; although it did not meet the criteria for diagnosis. Therefore, we treated the patient for stent-graft infection. Although graft infection after AUF treatment has not been reported, the risk of graft infection always exists. Most reports have identified *Staphylococcus spp.* and *Streptococcus spp.* as the most common causative organisms in endograft infection. Similarly, gramnegative organisms (eg, *Escherichia* coli and *Enterococcus*) have been identified and have previously been reported along with *Candida* [1]. This case is comparable with previous reports. The source of the infection could be cellulitis, or more likely a urinary tract infection, as the patient had a complex history of urinary tract treatment [5].

We assumed several potential reasons for the pseudoaneurysm formation:

- Graft infection occurred because the stent-graft was located next to the urinary tract, from where the infection spread to the normal artery. Focal perforation of the artery wall occurred, and a pseudoaneurysm was formed.
- 2) Stent graft edge injury accompanied by migration of the graft occurred and resulted in pseudoaneurysm formation.
- 3) Both 1) and 2) occurred.

The wall of a normal artery can be infected by the contiguous spread of a local infection, resulting in abscess formation, focal perforation, and pseudoaneurysm formation [11]. Pseudoaneurysm formation at the iliac artery resulting from stent-graft migration has been previously reported [12], and other reports have shown the possibility of stent-graft fracture due to posture [13] or compression of the abdominal wall [14]. Although the index patient did not have such history, there is still a possibility that edge injury resulted from migration or deformation of the stent-graft for some reason. Moreover, balloon-expandable stent-grafts could have migrated more commonly than self-expandable stent-grafts because of the lack of chronic outward force.

It is uncertain why the iliac artery occluded within a few days. However, the iliac artery could have been occluded when we performed angiography to detect the pseudoaneurysm because, at that time, a floating thrombus was already present. There are two potential reasons for thrombus formation. First, arterial flow limitation occurred because the arterial flow was stolen by the huge pseudoaneurysm. Second, the antithrombus medication was insufficient; the patient had taken only aspirin 100 mg/day as antiplatelet therapy since the stent-graft implantation.

Although there is no "gold standard" treatment strategy for stent graft infection, removing the infectious stent-graft is recommended [9]. However, the peri-procedural mortality rate with open surgery is 10%–40% [1' 15-17]; in contrast, the mortality rate of conservatively-treated patients is 30%–100% [6-9]. Extension with further stent-grafts was used in salvage situations to prevent pseudoaneurysm rupture. A previous study reported that this method had temporary effects [18]. In the index case, we considered open surgery unfavorable because the patient was elderly and very frail. Moreover, the blood culture became negative for MRSA on antibiotic therapy alone. Hence, along with appropriate antibiotics, we implanted an additional stent-graft to prevent bleeding and rupture of the pseudoaneurysm. In this case, the implantation of a new stent graft became unnecessary as the iliac artery spontaneously became occluded, and blood flow to the pseudoaneurysm disappeared. Although ischemia was present in the right limb, blood perfusion was confirmed, and the patient did not develop symptoms of limb ischemia. Therefore, we believe that invasive interventions were not mandatory in this case.

There is currently no consensus or data recommending an appropriate duration of antibiotic therapy [17, 19]. In this case, we believe that antibiotics should be continued for the rest of the patient's life because the infected stent graft was not explanted.

In conclusion, infection and pseudoaneurysm formation can occur following the implantation of a stent-graft. Patients who are treated with endovascular stent-grafts, especially those at higher risk of infection, should be followed up carefully.

Patient Consent

The patient provided written consent for the publication of this case and any accompanying images. Our institution does not require ethical approval for reporting individual cases or case series.

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