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Case Report

Inferior Vena Cava Occlusion and Ilio-Iliac Arteriovenous Fistula Caused by Tumor Invasion: A Case Report

Yukinori Shinoda, MD, Tetsuya Watanabe, MD, PhD, Tomoko Minamisaka, MD, Hidetada Fukuoka, MD, Hirooki Inui, MD, Keisuke Ueno, MD, Soki Inoue, MD, Kentaro Mine, MD, and Shiro Hoshida, MD, PhD

An 80-year-old woman presented with general fatigue and leg edema for several months. Ultrasonography and contrast-enhanced computed tomographic angiography revealed inferior vena cava thrombosis, ilio-iliac arteriovenous fistula (AVF), and iliac artery pseudoaneurysm. Furthermore, malignant cells were observed in the aspirated thrombus. Although thrombus aspiration and anticoagulant therapy were unsuccessful in reducing the thrombotic mass and alleviating her symptoms, endovascular therapy for AVF and pseudoaneurysm improved her leg edema without recurrence or any endoleak. These findings highlight that endovascular therapy can be effective in older adults with cancer because of its low invasiveness.

Keywords: deep vein thrombosis, ilio-iliac arteriovenous fistula, iliac artery aneurysm

Introduction

Renal cell carcinoma that has metastasized at the time of diagnosis accounts for 25%–30% of all renal cancer cases, with a 2-year survival rate less than 20%.[1] Small renal cell carcinoma is generally considered to have a good prognosis, even though 1.1%–6.2% of renal cell carcinomas with a diameter of 3.0 cm or less are associated with distant metastases and poor prognosis.[2,3] Furthermore, many patients experience complications because of inferior vena cava (IVC) thrombi,[4] and vascular invasion is an independent predictor of prognosis in patients with renal cell carcinoma.[5]

Arteriovenous fistulas (AVFs) are usually caused by trauma or iatrogenic injury,[6] though some research has suggested that many AVFs are caused by arterial pseudoaneurysms.[7] AVFs associated with aneurysms of the abdominal aorta or iliac artery are rare, accounting for 2%–4% of cases in patients with arterial rupture and 0.2%–1.3% of cases in those without.[8] AVF is often accompanied by back pain, high-output heart failure, and abdominal bruit; however, diagnosis may be delayed because of nonspecific symptoms.[9] In the present report, a rare case of IVC thromboembolism and ilio-iliac AVF with iliac pseudoaneurysm, diagnosed on the basis of the patient's leg edema has been discussed.

Case Report

The patient was an 80-year-old woman who had been receiving medical therapy for hypertension and dyslipidemia. Before visiting our hospital, she experienced gradual worsening of her leg edema and general fatigue over several months. Although her vital signs were stable, she had abdominal bruits below the navel and pitting edema below both thighs. Laboratory analysis revealed mild anemia (hemoglobin, 10.8 g/dL) and high levels of C-reactive protein (3.2 mg/dL) and D-dimer (16.5 μg/mL). Venous ultrasound examination revealed a thrombus-like echo in her IVC below the renal veins, and she was admitted to our hospital for examination for deep vein thrombosis. Although echocardiography revealed no significant abnormalities, contrast-enhanced computed tomography (CT) angiography revealed sub-massive pulmonary thromboembolism (PTE) in the central portion of both pulmonary arteries. Although the renal veins were intact, the IVC was enlarged, and we observed a lack of contrast just below

Department of Cardiovascular Medicine, Yao Municipal Hospital, Yao, Osaka, Japan

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Corresponding author: Yukinori Shinoda, MD, PhD. Department of Cardiovascular Medicine, Yao Municipal Hospital, 1-3-1 Ryuge-cho, Yao, Osaka 581-0069, Japan
Tel: +81-72-922-0881, Fax: +81-72-924-4820
E-mail: Yshinoda1031@gmail.com

the renal veins, as with sonographic venography. An ilio-iliac AVF of approximately 2 mm was observed between the right common iliac artery and the left common iliac vein. Pseudoaneurysms (17×12 mm) were detected just distal and to the left of the fistula (**Fig. 1**). The veins of the pelvic wall, abdominal wall, those around the vertebral body, and the ovary had expanded to form a collateral circulation. Based on these findings, she was diagnosed with PTE, IVC thrombosis, right ilio-iliac AVF, and right common iliac artery aneurysm.

First, in order to prevent the recurrence of PTE, a DENALI IVC filter (C. R. Bard, Inc., New Providence, NJ, USA) was implanted at the right internal jugular vein above the renal vein on the day of admission, as the IVC was occluded from the lower part of the spout of the renal veins. The contrast from the abdominal aorta flowed from the right common iliac artery to the left iliac vein, to the pelvic vein, and finally to the IVC. The mean blood pressure (BP) of the left and right common femoral veins increased to 52 and 37 mmHg, respectively. In addition, the BP of the abdominal aorta was 159/60 mmHg, and the mean IVC pressure of the upper portion from the spout of the renal veins was 17 mmHg.

The red thrombus was obtained via aspiration, which was repeated three times. We then placed the fountain catheter into the thrombus, and catheter-directed thrombolysis (CDT) was performed using urokinase for 5 days (240,000 units per day). Following CDT, we again performed CT angiography, which revealed that the IVC remained occluded. Although we performed balloon angioplasty using SABER6.0*150 mm (Cardinal Health Inc., Dublin, USA), the vessel immediately occluded again. Therefore, we prescribed a direct oral anticoagulant agent (apixaban, 5 mg, b.i.d.). Although results for all tumor markers (carbohydrate antigen 19-9 [CA19-9], α-fetoprotein [AFP], and carcinoembryonic antigen [CEA]) were negative, pathological findings of the thrombus obtained during endovascular therapy (EVT) led us to suspect renal cell carcinoma (**Fig. 2**).

Blood flow into the iliac veins through the ilio-iliac AVF took the pelvic collateral route due to IVC occlusion, leading to high venous pressure in the lower extremities and leg edema. We attempted to close the ilio-iliac AVF to reduce venous pressure in the lower extremities. Because the patient was suspected to have cancer, EVT was performed to close the AVF on the 15th day of admission. A pigtail catheter was delivered from the left radial artery to the abdominal aorta, following which angiography was performed. We inserted the sheaths into both common femoral arteries (CFAs). We observed the vessel around the AVF via intravascular ultrasound of the right CFA. The AVF was located at the middle of the right common iliac artery (CIA). The diameter and the length of the target vessel

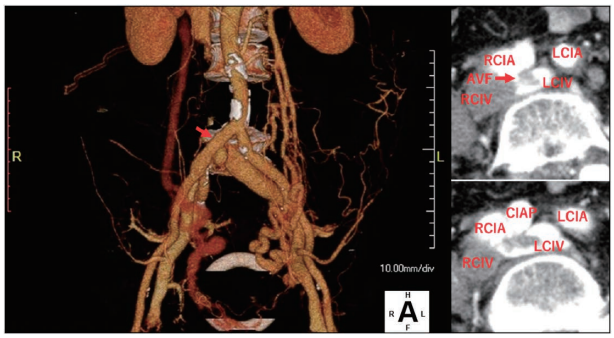

Fig. 1 Contrast-enhanced computed tomography revealed an ilio-iliac fistula between the right common iliac artery and left common iliac vein. An aneurysm was observed just internal to the AVF.
RCIA: right common iliac artery; LCIA: left common iliac artery; RCIV: right common iliac vein; LCIV: left common iliac vein; AVF: arteriovenous fistula; CIAP: common iliac artery pseudoaneurysm

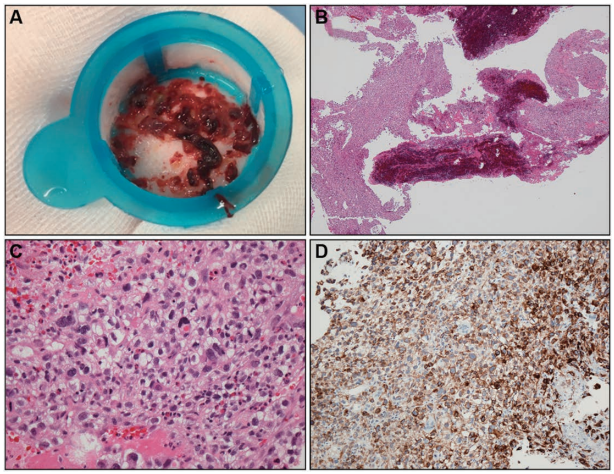

Fig. 2 (**A**) Clot obtained from the inferior vena cava thrombus. (**B**) Hematoxylin-eosin (HE) stain (×4), (**C**) HE stain (×40), (**D**) Immunohistochemistry stain (×20) (CAM5.2 positive).

were 12 and 40 mm, respectively. We implanted a stent graft (Viabahn 13×50 mm^2, W. L. Gore & Associates, Inc., Newark, NJ, USA) in the right CIA. The intravascular ultrasound (IVUS) from the left CFA revealed that the ostium of the left CIA had narrowed because of protrusion of the proximal edge of the stent graft into the abdominal aorta. Therefore, we delivered a balloon-type expandable stent (Assurant Cobalt 10×40 mm^2, Medtronic Vascular, Inc., Dublin, Ireland) from the left CFA, which was implanted in the left CIA. The proximal edge of the expandable stent was located at the same position as the edge of the stent graft. We then dilated both proximal ends of the stents using the kissing balloon technique. The AVF and aneurysm had disappeared following this procedure, and the final angiography examination revealed no leakage flow (**Fig. 3**).

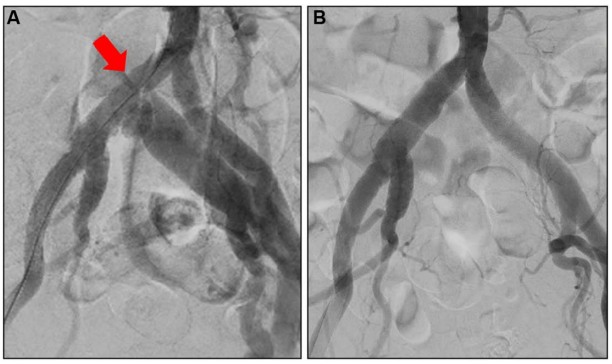

Fig. 3 (**A**) The contrast passed through the arteriovenous fistula (AVF) and flowed into the iliac vein. (**B**) The AVF, aneurysm, and iliac vein disappeared following endovascular therapy.

The patient was discharged from the hospital on day 21. By the first outpatient visit after discharge, her leg edema had mostly resolved. CT angiography and echo venography revealed disappearance of the AVF and aneurysm regression. No tumor was found in the kidney, and the renal vein was not occluded by the tumor or thrombus according to the CT images. However, the IVC filter was permanently indwelling because the IVC thrombus was enlarged after anticoagulant therapy. Multiple bone metastases were suspected within the pelvis. Since positron emission tomography–CT did not suggest a site of origin, the patient was diagnosed with cancer of unknown primary origin. She was then treated via chemoradiotherapy. We performed the CT several times during chemotherapy, and it was confirmed that the thickening of the vein wall at the IVC bifurcation had been regressed by chemotherapy. One year after the treatment, IVC thrombosis was enlarged, no recurrence of the AVF or aneurysm had been present.

Discussion

In the present report, we discussed the case of a patient with IVC thrombosis, pseudoaneurysm of the CIA, and ilio-iliac AVF. IVC thrombosis and ilio-iliac AVF may have been related to malignant cell invasion of the vascular bed.

Renal cell carcinoma tends to invade the venous system and metastasize to remote organs. In the present case, substantial growth of the primary tumor was not observed, and the IVC was occluded due to tumor invasion. However, due to the development of a collateral circulation involving the pelvic vein, leg edema was not immediately apparent. The CIA aneurysm may have formed at this location due to arteriosclerosis. The diameter of the aneurysm was approximately 17 mm, and the risk of rupture or penetration was extremely low. However, when the adjacent common iliac vein expanded due to the increase in venous pressure resulting from the IVC thrombus, AVF occurred, and venous tumor infiltration may have compressed the aneurysm. Venous blood flow and venous pressure rapidly increased, leading to an inability of the collateral circulation to compensate and subsequent leg edema. This also explains why leg edema improved following AVF treatment. Our patient did not experience high-output heart failure due to the IVC thrombus, instead presenting with leg edema influenced by venous retention in the lower extremities.

We selected EVT with a stent graft for the treatment of ilio-iliac AVF, which was thought to have caused the patient's leg edema. Recently, stent grafts have been increasingly used in the treatment of aortic aneurysms. Even in the iliac artery region, if the patient's vital signs are stable, EVT with stent grafting is superior to open surgery with regard to the duration of the operation and extent of bleeding.[10] In the present case, the size of the aneurysm was 17×12 mm and the diameter of the AVF was 2 mm, both relatively small and far enough from the ostium of the CIA and the bifurcation of the internal or external iliac arteries. In some cases, the internal iliac artery is embolized during the treatment of iliac artery pseudoaneurysms to prevent endoleak. However, embolization can cause ischemia within the pelvic organs. Given the small size of the iliac artery pseudoaneurysm, we chose to protect the internal iliac artery in our patient. Therefore, the stent graft was inserted into the abdominal aorta, narrowing the ostium of the left CIA. We preserved the ostium of the left CIA by implanting a balloon-expandable stent. Open surgery is recommended if enlargement of the aneurysms or endoleak is detected after implanting the stent graft.[10] CT examination after discharge revealed no endoleaks as well as a reduction in the size of the aneurysm. The patient has been followed up without additional invasive therapy.

Conclusion

The patient in the present case exhibited IVC thrombosis and ilio-iliac AVF, which may have been related to tumor invasion, causing lower leg edema. EVT with stent grafting successfully treated the AVF and reduced lower leg edema.

Disclosure Statement

The authors declare no conflicts of interest.

Author Contributions

Study conception: YS
Writing: YS
Critical review and revision: all authors
Final approval of the article: all authors

Accountability for all aspects of the work: all authors

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