

## A Case of Iliac Arteriovenous Fistula Presenting with Iliac Artery Aneurysm Preoperatively Diagnosed by Ultrasonography

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**Iliac arteriovenous fistula is a rare but severe complication of iliac artery aneurysm. We present a case of iliac arteriovenous fistula concomitant with iliac artery aneurysm, which was preoperatively diagnosed by ultrasonography (USG) and successfully treated with emergent surgery. An 84-year-old female admitted to our hospital complaining of a sudden onset of right leg edema and dyspnea. Physical examination revealed pansystolic murmur at the right inguinal region. A chest X-ray showed enhanced pulmonary vascular shadow and bilateral pleural effusion with cardiomegaly. USG of the right lower abdomen revealed an arteriovenous fistula between the right iliac artery and vein concomitant with the iliac artery aneurysm. An emergent surgery was performed, and the fistula was directly closed within the aneurysm. To reduce bleeding through the fistula during surgery, we placed fingers inside the aneurysm and compressed the iliac vein just after the aneurysmal sac was opened. The postoperative course was satisfactory. (Ann Thorac Cardiovasc Surg 2009; 15: 133–136)**

**Key words:** arteriovenous fistula, iliac artery aneurysm, emergent surgery, ultrasonography

### Introduction

An arteriovenous fistula is an uncommon complication presenting in 0.2%–1.3% of abdominal aortic aneurysms.<sup>1)</sup> The operative mortality is reported as 6%–40%.<sup>2–4)</sup> To reduce the mortality, preoperative diagnosis and controlling the bleeding during surgery are important. Diagnosis is correctly made in only 50% of arteriovenous fistulas before surgery.<sup>5)</sup> We report a case of iliac arteriovenous fistula complicated by an iliac artery aneurysm, which was preoperatively diagnosed by ultrasonography (USG)

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Received January 8, 2008; accepted for publication March 10, 2008

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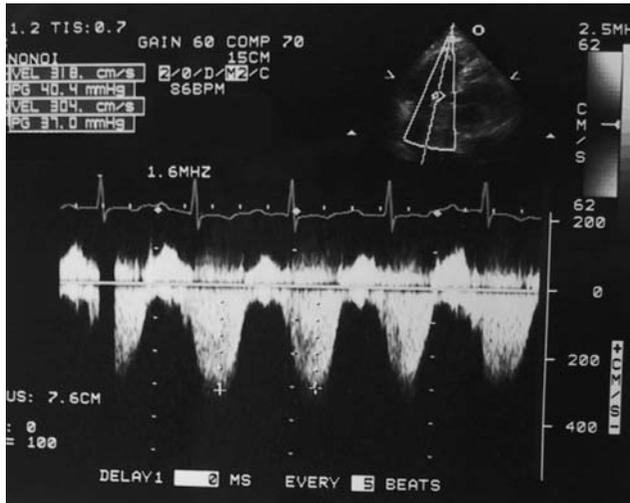
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and successfully treated with fistula closure and iliac artery replacement.

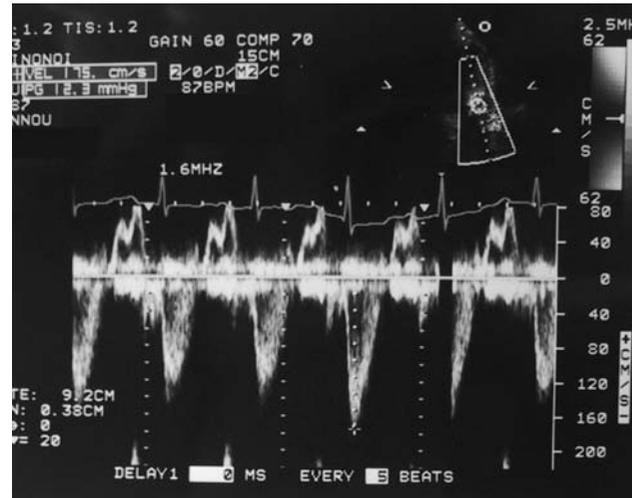
### Case Report

An 84-year-old female was admitted to our hospital complaining of a sudden onset of right leg edema and dyspnea. Her medical history revealed 20 years of hypertension.

Physical examination revealed a pansystolic murmur at the right inguinal region. The right leg, thigh, and foot showed significant edema, whereas no edema was observed on the contralateral side. Heart rate was 80/m; blood pressure was 127/61 mmHg, and an electrocardiogram showed an incomplete right bundle branch block and a flattened T wave in I, aVL, II, and III leads. A chest X-ray showed enhanced pulmonary vascular shadow and bilateral pleural effusion with cardiomegaly. Ultrasonic cardiography revealed pulmonary hypertension with moderate tricuspid regurgitation, though the left ventricle was normally contracting and ejection frac-



**Fig. 1.** Ultrasonography in the right lower abdomen. A monophasic flow was observed in the internal iliac artery.



**Fig. 2.** Ultrasonography in the right lower abdomen showed an arterIALIZED waveform in the right common iliac vein.

tion was 71%. A USG of the right lower abdomen showed a right iliac artery aneurysm with a diameter of 57 × 52 mm. A monophasic flow in the iliac artery was observed (Fig. 1). In the common iliac vein, an arterIALIZED shunt flow (Fig. 2) and a mosaic color flow were observed. Retrograde flow was also observed in veins below the femoral vein. These findings suggested an arteriovenous fistula between the right iliac artery and vein concomitant with an iliac artery aneurysm. A CT scan revealed a bilateral common iliac artery aneurysm (CIAA) and an internal iliac artery aneurysm (IIAA). The diameter of the right and left IIAA was 54 and 42 mm, respectively. Contrast enhancement showed early enhancement of the right common iliac vein, suggesting an arteriovenous fistula between the right internal iliac artery and common iliac vein (Fig. 3). The inferior vena cava and hepatic vein was dilated because of increased blood return caused by the shunt flow. Laboratory data indicated mild liver and renal dysfunctions. The B-type natriuretic peptide was 733 pg/ml.

Emergent surgery was performed. The abdomen was opened through a long midline incision. Significant subcutaneous edema and 500 ml of ascites were observed. The duodenum was mobilized and the retroperitoneum incised. The diameter of abdominal aorta was 23 mm, and no aneurysm was observed. The diameters of right and left IIAAs were 50 and 40 mm, respectively, and thrill was palpated above the right CIAA. After intravenous heparin administration, the aorta was clamped proximally at 3 cm above and distally 2 cm below the



**Fig. 3.** CT with contrast enhancement showed early enhancement of the right common iliac vein.

inferior mesenteric artery. The aorta was resected, and a Dacron graft anastomosed. The left CIAA and IIAA sacs were then opened. The orifice of IIAA was closed inside the aneurysm. The right graft limb was anastomosed to the right external iliac artery. After the left lower extremity was reperfused and an assistant was ready for controlling bleeding through the fistula, the right CIAA sac was opened. The assistant inserted the forefinger into the aneurysm and compressed the aneurysmal wall toward the common iliac vein. Perforation was found on the lateral wall of IIAA at 1 cm distal of the internal and external iliac bifurcation and sized 6 mm diameter. The

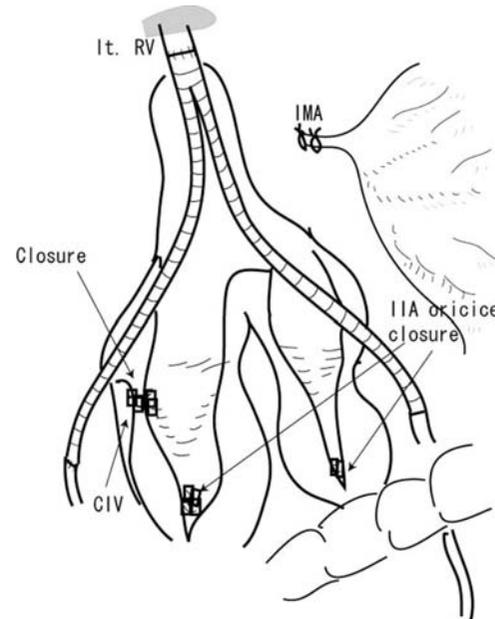
perforation was oversewn using the aneurysmal wall. After the IAA sac was fully opened, the orifice of IAA was closed in the same manner as the left side. The right external iliac artery was then resected and the right graft limb anastomosed (Fig. 4). We measured pressure of the inferior mesenteric artery by inserting a 22 gauge cannula through the orifice to compare with the radial artery pressure. The systolic pressures of the inferior mesenteric and radial arteries were 48 and 86 mmHg, respectively, and the inferior mesenteric artery was ligated. The operative time was 321 minutes, and total blood loss was 2,180 ml.

The postoperative course was uneventful and satisfactory. She started drinking and walking 4 days after surgery. No signs of bowel ischemia were observed. She was discharged from the hospital 24 days after surgery and is now experiencing normal daily life, though her right foot edema remained 3 weeks after surgery. CT scan revealed that the left external iliac vein was occluded by thrombus.

## Discussion

Iliac arteriovenous fistula is a rare complication of iliac artery aneurysm. Arteriovenous fistula was presented in 0.2%–1.3% of abdominal aortic aneurysm and in 3.0%–4.0% of its rupture.<sup>1)</sup> Duong and Atkinson reported that 14 cases of arteriovenous fistula were found in 2,249 abdominal aortic aneurysms that were surgically treated in their institution.<sup>5)</sup> In those 14, only 2 were ilio-iliac fistula, 11 were aortocaval, and 1 was aortoiliac. In other literature, only a few cases of iliac arteriovenous fistula were reported.<sup>6–8)</sup> Solitary iliac aneurysm, which is without aortic aneurysm, is also rare, and its incidence is less than 1%.<sup>9)</sup> Our case was ilio-iliac arteriovenous fistula presenting with isolated iliac artery aneurysm and was considered uncommon.

Preoperative diagnosis is critical to reduce blood loss during surgery and mortality of arteriovenous fistula. Duong and Atkinson described that preoperative diagnosis was correctly made in only 50% of arteriovenous fistula with aortoiliac aneurysm.<sup>5)</sup> Cinara et al. reported 26 cases of aortocaval fistula, and they made a correct diagnosis in 62% preoperatively. In these 26 cases, 5 operative deaths were observed and were involved in incorrect diagnoses before surgery.<sup>3)</sup> In our case, we prepared for bleeding through the fistula before we opened the aneurysmal sac because of the preoperative diagnosis. Correct preoperative diagnosis reduced



**Fig. 4.** Operative schema.

A Y-shaped Dacron graft was used for reconstruction. The orifices of the left and right internal iliac arteries (IIAs) were closed inside the aneurysm. Perforation found on the lateral wall of the IIA was oversewn using the aneurysmal wall. IMA, inferior mesenteric artery; It. RV, left renal vein; CIV, left common iliac vein.

unexpected blood loss during the surgery.

The diagnosis in our case was made by USG, which was beneficial for prompt treatment and subsequent clinical course. Pinheiro et al. reported a case of ilio-iliac arteriovenous fistula diagnosed by USG. However, they described that USG was unreliable for detection in the abdominal region because of bowel gas.<sup>10)</sup> Another case was reported by Huang et al.<sup>11)</sup> A fistula tract was visualized between the right CIAA and the iliac vein. A pulsed Doppler study showed a monophasic waveform in the right common iliac artery arousing speculation that the iliac artery had a shunt flow toward the low resistant vein. Continuous turbulent flow from the iliac aneurysm to the vein was presented as a mosaic flow in duplex color study. Li et al. proposed diagnosis criteria to locate arteriovenous fistula by USG.<sup>12)</sup> The major diagnostic criteria are junction of low- and high-resistance flows in the supplying artery, a high-velocity arterialized waveform in the draining vein, and a turbulent high-velocity flow spectrum at the junction of the artery and the vein. The minor diagnostic criteria are direct communication between the involved artery and vein, significant change in the diameter of the supplying artery, a focal point of

venous dilatation, and a focal perivascular color artifact. In our case, the patient was not obese, and the bowel gas did not interrupt ultrasound during the examination in the lower abdomen. A Doppler study showed the monophasic waveform in the iliac artery and the arterialized shunt flow in the iliac vein. In the duplex scan, the mosaic color flow was observed in the iliac vein. These findings are in accord with their criteria.

It is also important to suspect the presence of arteriovenous fistula from the patient's symptom and sign. The patient may present a classical triad of high-output cardiac failure, pulsatile abdominal mass with bruit and unilateral ischemia or venous congestion.<sup>13)</sup> In our case, the patient complained of a sudden onset of right leg edema and dyspnea. Pansystolic murmur at the right inguinal region and the right leg edema was observed. We performed UCG in the right lower abdomen on the same day the patient was admitted because the arteriovenous fistula was strongly suspected in the outpatient clinic. We would recommend that physicians proceed to examine the patient with suspicion of the arteriovenous fistula when the patient shows a triad of high-output cardiac failure, pulsatile abdominal mass with bruit, and unilateral ischemia or venous congestion.

Another critical concern is how to control the bleeding through the fistula when the aneurysm sac is opened during surgery. This is done by direct compression of the vein and balloon catheters inserted into the vein. Rajmohan reported that the bleeding from the fistula was controlled by compressing the vein with swab sticks.<sup>14)</sup> Naito et al. inserted balloon catheters into the vein proximally and distally to the fistula prior to surgery and thus reduced the bleeding.<sup>15)</sup> We placed fingers inside the IAA sac and compressed the iliac vein over the lateral wall of IAA to reduce the bleeding through the fistula. The aneurysmal and venous walls were firmly attached and difficult to divide. It is important to manipulate the vein over the aneurysmal wall.

In conclusion, we experienced a case of ilio-iliac arteriovenous fistula concomitant with IAA. The patient was preoperatively diagnosed by USG and successfully treated by emergent surgery.

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