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# Successful surgical repair of an ilio-iliac arteriovenous fistula associated with a ruptured common iliac artery aneurysm



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## ABSTRACT

**INTRODUCTION:** We describe the case of an 86-year-old man with an ilio-iliac arteriovenous fistula (AVF) resulting from a ruptured aneurysm. This condition rarely occurs, has a high mortality rate, and was successfully treated via surgery.

**PRESENTATION OF CASE:** The patient presented with a temporary loss of consciousness and left leg edema. A pulsatile abdominal mass with vascular murmur and thrill was detected. Enhanced computed tomography showed abdominal aortic and iliac aneurysms with left common iliac vein occlusion, and the left external iliac vein was easily seen through the AVF. We directly sutured the AVF and performed aneurysmectomy and prosthetic graft replacement. During surgery, placement of occlusive balloon catheters through the AVF minimized intraoperative bleeding. The patient recovered uneventfully, and swelling of the left leg was immediately reduced after surgery.

**DISCUSSION:** Although rare, AVFs can be life-threatening, and urgent treatment and intensive care are occasionally needed. Surgical management of AVF requires a definitive preoperative diagnosis and control of venous bleeding during surgery. Fulfilling these major requirements can potentially reduce morbidity and mortality in patients with AVFs. Interestingly, there was no sign of high-output heart failure throughout the treatment course due to compression of the aneurysm and consequent blood flow failure to the left iliac vein.

**CONCLUSION:** Using the balloon occlusion technique, we were able to minimize blood loss during open repair. Use of multiple imaging modalities facilitates correct preoperative diagnosis and consequently improves surgical outcome.

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## 1. Introduction

Ilio-iliac arteriovenous fistula (AVF) is a rare disease that occurs in less than 1% of all common iliac artery aneurysms (CIAAs) and in 27% of ruptured aneurysms [1]. Surgical repair of an AVF resulting from a ruptured abdominal aortic aneurysm (AAA) was first described by Cooley in 1955 [2], and several similar cases have since been reported. Nevertheless, perioperative mortality in patients with AAAs with AVFs is still as high as 67% in some series [3,4]. Preoperative diagnosis of AVF is difficult, and control of venous bleeding during open surgery can affect surgical outcome. Here we report a case of a contained rupture of a left CIAA with an ilio-iliac

AVF that was diagnosed preoperatively and successfully treated via surgery.

## 2. Presentation of case

An 86-year-old man was admitted to a local hospital because of a temporary loss of consciousness and left leg edema. His medical history was unremarkable. Plain computed tomography (CT) showed enlargement of the abdominal aorta and left common iliac artery (CIA), and he was transferred to our institution for further evaluation and treatment.

Upon admission, the patient's blood pressure was 130/76 mmHg, and his heart rate was 88 beats/min. A pulsatile abdominal mass with vascular murmur and thrill was noted. The left lower leg was considerably swollen compared with the lower right leg (left: 37.0 cm, right: 34.4 cm). No clinical signs of high-output heart failure, including hepatomegaly, jugular vein dilatation, and pleural effusion, were observed. Blood test results (hemoglobin, 10.4 g/dL; hematocrit, 30.9%) indicated anemia. Arterial phase CT (GE Optima CT 660, GE Healthcare, Tokyo,

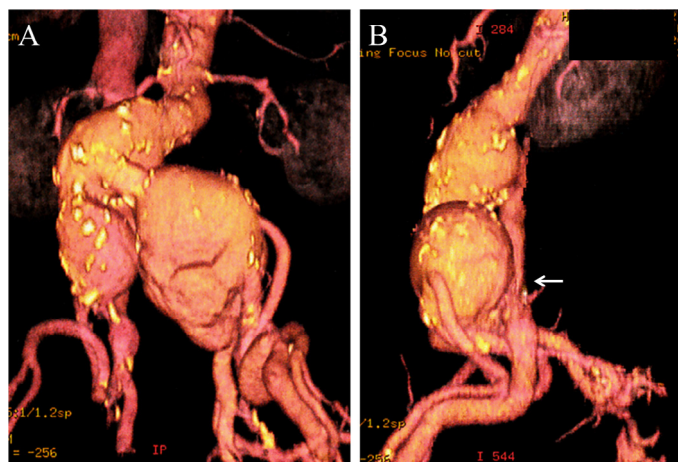
*Abbreviations:* AVF, arteriovenous fistula; CIAA, common iliac artery aneurysm; AAA, abdominal aortic aneurysm; CT, computed tomography; CIA, common iliac artery; CIV, common iliac vein.

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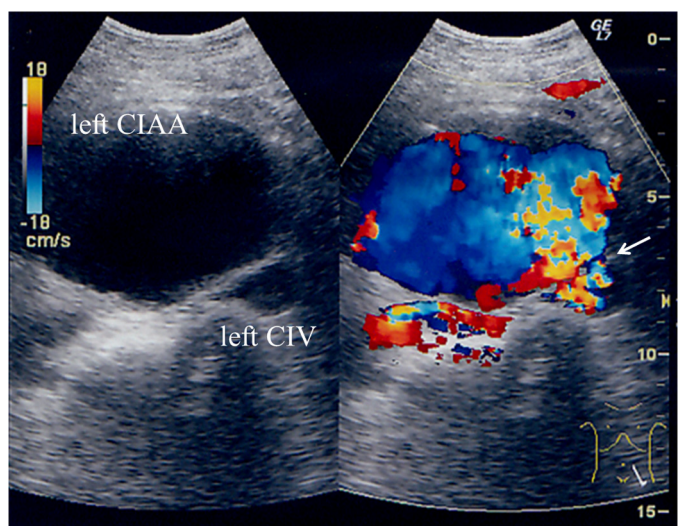
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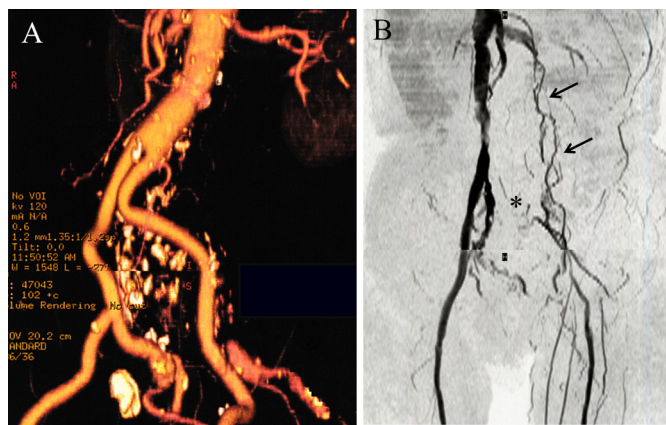
**Fig. 1.** Preoperative contrast-enhanced computed tomography three-dimensional reconstruction shows a 60-mm abdominal aortic aneurysm (A) and a 70-mm left common iliac artery aneurysm with an ilio-iliac AVF originating in the left common iliac artery (arrow) (B).



**Fig. 2.** Abdominal ultrasonography shows the shunt flow from the left common iliac artery aneurysm to the left common iliac vein (arrow).

Japan) of the abdominal and pelvic vessels using a contrast agent (Omnipaque, 300 mg/mL; Daiichi Sankyo Pharma, Tokyo, Japan) showed an infrarenal AAA (maximum diameter, 60 mm) and a left CIAA (70 mm) complicated by a pressed left common iliac vein (CIV). Early appearance of contrast medium in the dilated left CIV was notable and indicated the presence of an ilio-iliac AVF (Fig. 1). An abdominal echo revealed a mosaic flow, suggesting a shunt between the left CIA and CIV (Fig. 2). The clinical presentation and imaging findings led to the diagnosis of a contained rupture of the left CIAA exploding into the left CIV. The patient underwent elective surgery.

A midline laparotomy exposed the infrarenal AAA and left CIAA. After proximal and distal control of the aneurysm was obtained, the aneurysm was opened longitudinally, and intramural thrombi were removed. Opening the left CIAA revealed the AVF on the posterior wall of the aneurysm. Venous bleeding was controlled via balloon occlusion (TMP balloon catheter 9 Fr, Tokai Medical Products Inc., Aichi, Japan) through the fistula. The AVF was approximately 10 mm in diameter and was closed directly via continuous suture using 4-0 monofilament, non-absorbable polypropylene thread (Ethicon Inc., Somerville, NJ, USA). A Dacron bifurcated graft



**Fig. 3.** (A) Postoperative contrast-enhanced computed tomography three-dimensional reconstruction shows a patent graft and the absence of the arteriovenous fistula. (B) Magnetic resonance venography shows left iliac vein occlusion (asterisk) and significant collateral circulation (arrows).

[INTERGARD 16 × 8 mm, InterVascular S.A. (MaquetCardiovascular, La Ciotat, France)] was anastomosed proximally to the infrarenal aorta and distally to the right CIA and the left external iliac artery. The left internal iliac artery was closed directly, and the inferior mesenteric artery was reconstructed by anastomosing the left leg of the graft.

The patient recovered uneventfully, and swelling of the left lower extremity quickly disappeared. Postoperative contrast-enhanced CT revealed a patent graft and absence of the AVF (Fig. 3A); however, magnetic resonance venography showed left iliac vein occlusion and significant collateral circulation (Fig. 3B). The patient was discharged on postoperative day 29.

### 3. Discussion

Abdominal AVFs are anomalous passageways between the abdominal aorta, iliac artery, or renal artery and the inferior vena cava, iliac vein, or renal vein. They result not only from aneurysmal diseases (primary cause), but also from iatrogenesis, malignancy, and trauma such as gunshot wounds (secondary causes) [5,6]. Interestingly, they can also develop in patients with tuberous sclerosis or Down syndrome [7,8]. Although rare, abdominal AVFs can be life-threatening, and intensive care management and urgent treatment consisting of open surgery or endovascular repair are occasionally required.

In their review of case reports, McAuley et al [9] identified three symptoms consistently associated with the presence of an AVF: (1) high-output cardiac failure with a precipitous onset, (2) a pulsatile abdominal mass accompanied by a thrill and a bruit, and (3) unilateral lower-extremity ischemia or venous engorgement. However, this classic triad is found in only 50–80% of all AVF cases [10]. A variety of symptoms appear depending on the size and location of the AVF, time after the rupture, cardiopulmonary function, and bleeding into the retroperitoneum [3]. Consequently, the correct diagnosis is made in only 37–52% of cases before surgery [11]. Several case reports recommend the use of CT or duplex ultrasound as a means of diagnosing AVFs [12,13]. Any combination of imaging modalities, such as contrast-enhanced CT and abdominal ultrasonography, as used in this case, are useful, especially for regional diagnosis of AVF. Definitive preoperative diagnosis has the potential to reduce morbidity and mortality in surgical management.

Unresolved issues in open repair, which was performed in our study, include the control of venous bleeding and the prevention of pulmonary embolisms caused by debris or air. In the present case, a balloon occlusion catheter was used to minimize intraoperative

bleeding from the fistula, as previously reported [12,14]. Bleeding from the AVF can usually be controlled via digital compression. However, we suggest that the balloon technique be considered as a stand-by procedure in open repair. It may be particularly useful when the AVF is large or the vessel wall is fragile.

Treatment of aneurysms with AVFs via endovascular repair has become increasingly more common in recent years. Compared with open repair, it is less invasive, and the risk of major blood loss during treatment is low. However, its long-term performance is still unknown. In a systematic review, the longest follow-up time after endovascular repair was 24 months, and the mean follow-up time was only 9 months [15]. Endovascular repair also carries the risk of type 2 endoleak due to venous bleeding through the AVF and consequent expansion of the aneurysm. Conclusions regarding this procedure require long-term follow-ups and randomized controlled trials with large case numbers. In the current case, the patient's anatomy was unsuitable for endovascular treatment, and technical difficulties due to AVF coil embolization were envisioned. If his anatomy had been suitable, endovascular repair would have been considered as an initial therapy in view of his age.

Notably, in the present case, there was no sign of high-output heart failure throughout the treatment course. Postoperative magnetic resonance venography indicated blood flow failure of the left iliac vein due to compression of the aneurysm. The presence of considerable collateral circulation suggests that the compression occurred many years ago. We conclude that the main cause of the swelling in the left leg was not due to left iliac vein occlusion but to an AVF, because the swelling was immediately reduced after surgery.

#### 4. Conclusion

We report the successful surgical treatment of an ilio-iliac AVF associated with a ruptured CIAA. Using the balloon occlusion technique, we were able to minimize blood loss during open repair. Definitive diagnosis of AVF is sometimes difficult, because subjective symptoms and objective findings can be caused by various conditions. Use of multiple imaging modalities facilitates correct preoperative diagnosis in early disease stages and consequently improves surgical outcome.

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None.

#### Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Author contributions

Makoto Iijima: study design, data collection, data analysis, and manuscript preparation.

Masakazu Kawasaki: data collection and critical input.

Yoshimitsu Ishibashi: data collection and critical input.

#### Conflict of interest

None of the authors have any conflict of interest.

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