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Case Report

Spontaneous ilio-iliac arteriovenous fistula: A rare complication of aorto-iliac aneurysm ☆

Fatima Zohra Benbrahim, MD, Majda Ankri, MD, Asmae Fariyou, MD, Omar El Aoufir, PhD, Fatima Zahra Laamrani, PhD, Jroundi Laila, PhD*

Department of Emergency Radiology, Ibn Sina University Hospital, Mohammed V University, Rabat, Morocco

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ABSTRACT

Ilio-iliac arteriovenous fistula is an unusual complication of aorto-iliac aneurysms that can occur spontaneously, traumatically or iatrogenically. The typical clinical presentation includes the characteristic triad of high-output heart failure, a pulsatile abdominal mass with unilateral limb ischemia, or signs of venous congestion. We describe a rare case of spontaneous rupture of an aortoiliac aneurysm into the left common iliac vein of a 65-year-old man, easily diagnosed by angiography. We highlight here the angiographic findings of the ilio-iliac fistula, which was the means of diagnosis in this presentation, especially in patients with atypical clinical features at the outset, and we report the difficulties in choosing the optimal vascular approach.

Introduction

Aneurysms of the abdominal aorta with or without involvement of the iliac arteries have a prevalence between 2 and 11% in the population over 65 years of age [1]. However, they represent 1 of the main causes of death in men over >65 years of age, if left untreated above a certain diameter [1].

Ilio-iliac arteriovenous fistula (AVF) between the iliac artery and the common iliac vein is a rare and serious complication, affecting only 1% of all abdominal aortic aneurysms [2]. Spontaneous rupture of the aneurysm is the most common cause of arteriovenous fistula [2]. These fistulas classically present as a triad comprising high-flow heart failure, a pulsatile abdominal mass with unilateral venous ischemia or congestion of the leg [3]. We present here a rare case of a spontaneous ilioiliac arteriovenous fistula complicating an aneurysm of the subrenal abdominal aorta and iliac arteries occurring in a 65-year-old man.

Case presentation

A 65-year-old man with a medical history of hypertension, hypercholesterolemia and chronic smoking who presented to

Abbreviations: AVF, Arteriovenous fistula; IVC, Inferior vena cava; CT, Computed tomography; MIP, Maximum Intensity Projection; IA, iliac artery; IV, iliac vein.

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* Corresponding author.
E-mail address: fzohrabenbrahim93@gmail.com (F.Z. Benbrahim).

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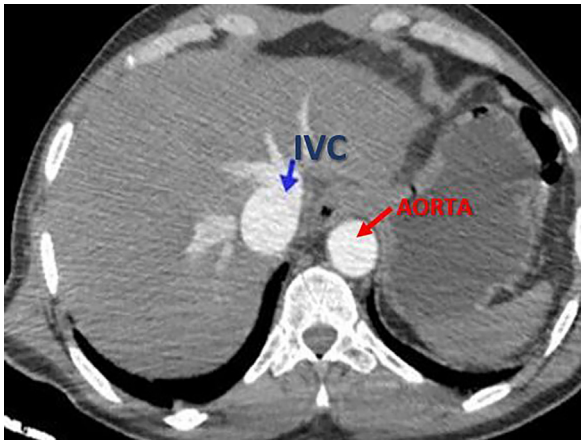

Fig. 1 – CT Angiographic at arterial phase in axial section showing dilatation with simultaneous opacification of IVC and aorta in arterial phase.

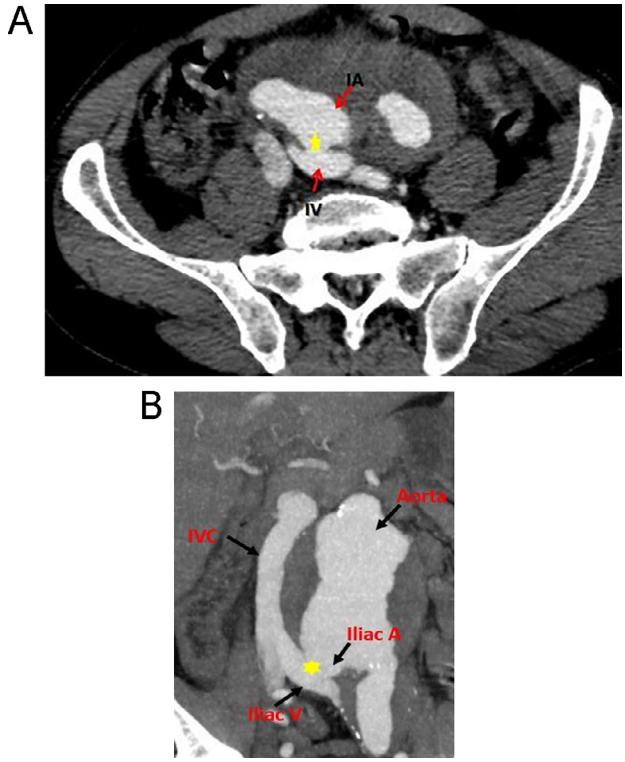

Fig. 2 – CT Angiographic images at arterial phase in axial section (A) and coronal MIP reconstruction (B) showing an aorto-biliac aneurysm with communication between the right common iliac artery and the left common iliac vein: arteriovenous fistula (yellow star). Note the retrograde enhancement of the IVC.

emergency department with acute periumbilical abdominal pain associated with edema of the right lower limb, which appeared enlarged big, pale, and cold. Interrogation was negative for any history of trauma or surgery. On admission, general examination found a patient in good general condition, apyretic, and hemodynamically stable, with blood pressure of 105 mmHg, heart rate of 105 beats/min, respiratory rate of 22 cycles/min, and oxygen saturation of 95%. Abdominal examination revealed a tender abdomen with a voluminous, pulsatile periumbilical mass, palpable thrill, and a systolic murmur upon auscultation, along with painful hepatomegaly.

Cardiovascular examination showed signs suggestive of right heart failure, including turgid jugular veins and oedema of the right lower limb. Vascular examination revealed the presence of femoral pulses, while distal pulses were difficult to detect.

Biological examinations showed normal levels of hemoglobin (12g/dL), platelets (265,000/mL) and white blood cells (8300/mL), with a slight rise in CRP: 5mg/dL (normal value of $<$ 0.3 mg/dL) and creatinemia at the upper limit: 12mg/dL. The rest of the liver function tests and ionogram were normal.

An emergency abdominal ultrasound with Doppler revealed an aneurysm of the sub-renal abdominal aorta extending to the primitive iliac arteries, partially thrombosed, measuring 90 $\times$ 98 mm. Additionally, there was dilatation of the inferior vena cava measuring 30 mm (anteroposterior diameter) with arterialized flow on pulsed Doppler, suggesting an arteriovenous fistula.

CT Angiography in the arterial phase (Figs. 1-3) revealed a fusiform aneurysm of the sub-renal abdominal aorta extending to the common iliac arteries bilaterally. It was partially thrombosed, located 16 mm from the renal arteries and measuring 95 $\times$ 103 $\times$ 145 mm (T x AP x Height). Additionally, there was evidence of a communication between the right primitive iliac artery and the left primitive iliac vein, situated precisely at the junction of the aorta and the right common iliac artery. This was accompanied by simultaneous opacification of the primitive iliac veins and the inferior vena cava in the arterial phase.

Therapeutic decision was surgical in consideration of the aneurysm's large size, extension to the primitive iliac arteries and infiltration of periaortic fat. Surgical intervention involved closing the fistula and replacing the affected vessels with an appropriate bi-femoral aorto-prosthesis.

The postoperative course was straightforward without complications with a favorable clinical outcome.

Discussion

The spontaneous rupture of the aneurysm into the adjacent venous system is the most common cause of arteriovenous fistulas involving the abdominal aorta or iliac arteries, accounting for around 80% of all reported cases [4,5]. Traumatic and iatrogenic causes are incriminated in approximately 20% of cases [6]. Most of these spontaneous aneurysms are atherosclerotic, although cases of arteriovenous fistulas from syphilitic or mycotic aneurysms have been reported [7], as well as aneurysmal lesions observed in Marfan syndrome [8], Ehlers-Danlos syndrome [9], or Takayasu arteritis [10]. In rare cases, tumors can lead to arteriovenous fistulas through erosion of adjacent arterial and venous structures [11].

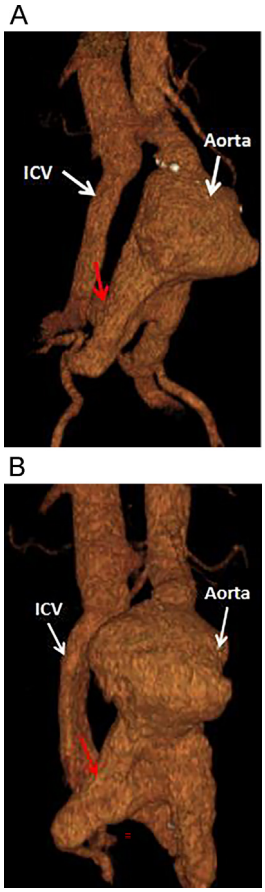

Fig. 3 – 3D reconstructions, showing aneurysmal dilatation of the sub-renal aorta and iliac arteries with visualization of the ilio-Iliac fistula (red arrow).

Ilio-iliac arteriovenous fistula is a rare complication of aortoiliac aneurysms, much less frequent than aortocaval fistula, with an incidence of less than 1% of all aneurysms and between 3% and 4% of ruptured aneurysms [2].

Typical symptoms of ilio-iliac arteriovenous fistula (AVF) include high-flow heart failure, abdominal pain, a pulsatile abdominal mass with murmurs and thrills, unilateral limb ischemia or signs of regional venous congestion such as lower-limb edema and hematuria [12]. Unusual clinical findings like dyspnea or hypotension are also possible. Gregoric et al. reported a case of acute renal failure associated with ilio-iliac AVF[13]. AVF can often be overlooked, especially in the presence of non-specific abdominal pain or signs of high-flow heart failure, which can have fatal consequences if left unrecognized. Brewster et al., in a 30-year retrospective study of 7 cases of iliac arteriovenous fistula, reported a latency period between the first symptoms and fistula diagnosis ranging from weeks to months, compared with the dramatic presentation of aortocaval fistula [14]. Our patient's presentation was more dramatic than usually seen in aorto-caval fistula cases, possibly due to the large size of the fistula.

The role of diagnostic imaging is crucial in the identification of the underlying pathology, in order to institute appropriate and rapid treatment. Traditionally, angiography has been used as a preoperative diagnostic means [14], but in recent decades and with advances in imaging technology, diagnosis by other methods has been reported.

B-mode ultrasound can represent the first-line examination, revealing aneurysmal dilatation with longitudinal extension into the renal and visceral arteries. Color Doppler shows a turbulent blood flow within the inferior vena cava (IVC) and provides a better assessment of the circulating lumen. The pulsed Doppler spectrum typically displays a systolic spike with an arterial flow diastolic component upstream of the arteriovenous fistula. However, direct visualization of the iliac fistula by pulsed Doppler has been described, but is generally unreliable, as this region is usually obstructed by bowel gas interposition [15].

Cheung et al. were the first to describe the use of dynamic CT with intravenous iodinated contrast agents injection to diagnose an iliac arteriovenous fistula [12]. Multi-detector CT has become the gold standard in imaging, permitting a rapid and highly accurate preoperative diagnosis to guide therapeutic management of AVF, particularly in cases with atypical clinical symptoms [16,17]. CT scan provides all the information required for therapeutic management, including aneurysm size, collar, longitudinal extension to the celiac trunk, and to the renal and iliac arteries, iliofemoral network condition, presence of arterial or venous anatomical variants, morphology of the aneurysmal lesion (wall, endoaneurysmal thrombus), and the periaurysmal environment. In addition, various computerized reconstruction techniques based on CT, such as volume reconstruction, maximum intensity projection and virtual angiography, can be used to localize AVFs [16].

Typical CT features include visualization of an aorto-iliac aneurysm, presence of retroperitoneal effusion, dilated IVC, and an early enhancement of the IVC, which displays a density similar to the aorta during arterial phase acquisition, due to shunting of vascular flow through the fistula. Normally, opacification of the infra-renal IVC typically occurs 60 seconds after maximal arterial enhancement, as most of the flow passes through the common iliac veins [18].

Until recent decades, open surgery was the recommended treatment for arteriovenous fistulas, carrying high mortality rate (20%-50%) [2,14,19]. Because the difficulty of approaching abdominopelvic vessels and venous hyper pressure, surgical repair of these lesions was associated with a high rate of bleeding and mortality, especially in hemodynamically unstable patients [20,21]. Additionaly, open surgery is also associated with risks of wound infection.

Endovascular treatment, introduced as a less invasive alternative since the early 1990s, has become a well-established option for managing ilio-caval arteriovenous fistulas (AVFs). This approach entails placing a covering stent to close the shunt while maintaining arterial and venous permeability [20,22]. However, the applicability of endovascular repair may be limited by factors such as the patient's comorbidities, anatomical considerations, and the availability of necessary equipment [2,23].

Endovascular treatment is particularly indicated for patients at high surgical risk [24]. However, in cases where obliterative atherosclerosis of the external iliac artery poses a risk of luminal obstruction, or in large aorto-iliac aneurysms

causing symptoms due to compression of surrounding tissues, open surgery remains the only alternative.

More recent hybrid techniques are also in use, involving initial coverage of the venous fistula by conventional stent grafting, followed by open surgical management of the aneurysm [25]. Endoleaks are the main complication after endovascular repair of aortic aneurysms, occurring in 45% of cases [26], and must be monitored regularly. Endovascular treatment remains associated with low mortality and bleeding compared with open surgery, in which mortality remains estimated at 5%-66% [27]. Additional retroperitoneal rupture of the aneurysm considerably increases mortality [26].

In our case, although endovascular treatment was proposed, it was not judged appropriate given the acute clinical presentation, the large size of the aorto-iliac aneurysm, the large ilio-iliac fistula, and the periaortic infiltration suggesting a pre-rupture.

In conclusion

Although ilio-iliac arteriovenous fistula is an unusual complication of aorto-Iliac aneurysm, it's important to recognize this condition and understand its clinical and hemodynamic aspects. The diagnosis should be suspected in any patient presenting with a pulsatile abdominal mass accompanied by signs of high-flow heart failure, arterial insufficiency or unilateral venous congestion. We highlight the crucial role of radiological diagnosis, particularly CT angiography, which serves as the gold standard when the clinical features of arteriovenous fistulas are atypical. Additionally, we acknowledge the challenges to choose the optimal vascular approach.

Patient consent

Written informed consent for the publication of this case report was obtained from the patient.

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