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Case Report

Endovascular wall-grafting for mycotic common iliac pseudo-aneurysm: Systemic mycoses [☆]

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ARTICLE INFO

Article history:

Received 20 April 2024

Revised 28 April 2024

Accepted 29 April 2024

Keywords:

Case report of an isolated common

iliac artery pseudoaneurysm

Iliac artery aneurysm

Candida albicans

Endovascular wall-grafting

ABSTRACT

Mycotic pseudoaneurysms of the iliac arteries are extremely rare and are caused by infection of the artery wall. It is difficult to diagnose early due to its silent manifestation.

We present a case of a 64-year-old man with an isolated left common iliac artery pseudoaneurysm caused by *Candida albicans* who presented with thrombophlebitis, abdominal pain, nausea and vomiting associated with fever, which was successfully treated with interposition grafting and antibiotic therapy.

We present this case to highlight that aneurysms and other vascular lesions can affect different arteries in the body and may therefore only be discovered during routine investigations of non-specific symptoms.

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Introduction

Unlike a true aneurysm, pseudoaneurysms do not have all three layers of the vessel wall. Since they are essentially a contained rupture, they have either adventitia or some media with only adventitia. As such, they are at high risk of rupture and when they do rupture, the mortality rate is as high as 50% [1].

The vast majority of isolated iliac artery aneurysms are asymptomatic and are discovered incidentally on imaging studies performed for other indications. Due to the deep pelvic location of these aneurysms, symptoms such as local visceral

or venous compression, neuropraxia or rupture may not occur until the aneurysms have reached significant size [2].

We report a case of a mycotic common iliac pseudoaneurysm caused by *Candida albicans* that was successfully treated with an endovascular graft.

Case report

A 64-year-old man with a history of diabetes mellitus, rheumatic disease on corticosteroid therapy and chronic tobacco use, no medical problems or hospitalizations and

[☆] Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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<https://doi.org/10.1016/j.radcr.2024.04.096>

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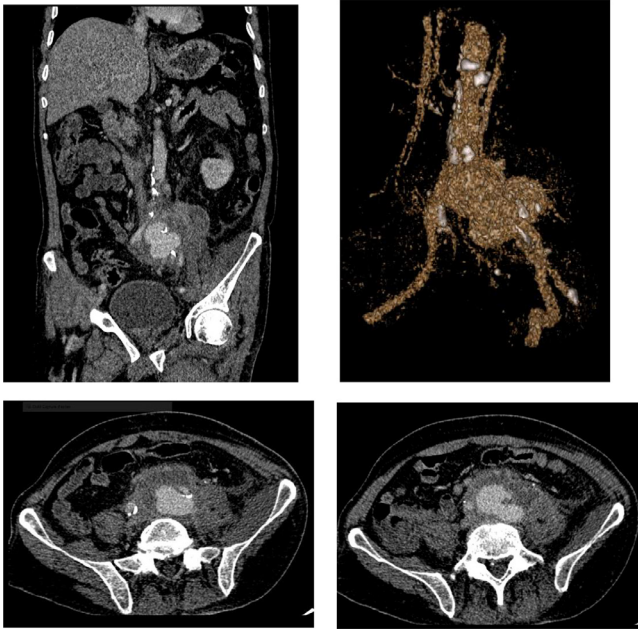


Fig. 1 – Contrast-enhanced computed tomography scan obtained at admission shows a 27 mm left common iliac artery pseudo-aneurysm with a 5 mm proximal neck, with an irregular lumen, which was contiguous with a left psoas muscle abscess, pelvic external iliac and left obturator adenopathy, and ipsilateral uretero-hydronephrosis.

no allergies, was admitted to the gastroenterology department because of abdominal pain, uncoercive vomiting, persistent hiccups and fever. On admission he appeared ill, his temperature was 38.2°C, pulse 100 beats per minute and blood pressure 130/80. Physical examination revealed oedema of the left lower limb extending to the thigh and chronic inguinal intertrigo. Leukocyte count was 20,000/mm³, neutrophil polynuclear count was 16,700/mm³, hemoglobin was 10.6, albumin was 25, C-reactive protein was 364; other laboratory tests were normal. An echo Doppler of the lower limbs showed a left femoro-popliteal thrombophlebitis, anticoagulation was prescribed. Gastro-duodenal fibroscopy revealed grade C esophagitis. An abdominal-pelvic computed tomography (CT) scan (Fig. 1) showed a left common iliac artery pseudoaneurysm with a bilobed aspect adjacent to a left psoas abscess, pelvic external iliac and left obturator adenopathy, and ipsilateral ureteral hydronephrosis.

The patient was started on intravenous amoxicillin-clavulanic acid with a remarkable reduction in inflammatory markers and anemia.

We opted for an endovascular approach (Figs. 2 and 3). Local anesthetic was infiltrated into both groins and both femoral arteries were accessed percutaneously (5 Fr sheath on the right, 9 Fr on the left). An Amplatz 0.035-inch wire was successfully introduced via the tortuous iliac artery. The pseudoaneurysm was identified. An 8-50 mm stent graft (wall graft) was advanced from the ipsilateral common femoral artery through a 9 Fr sheath and deployed across the ruptured lesion. As a proximal endoleak remained, an additional 8-50 mm

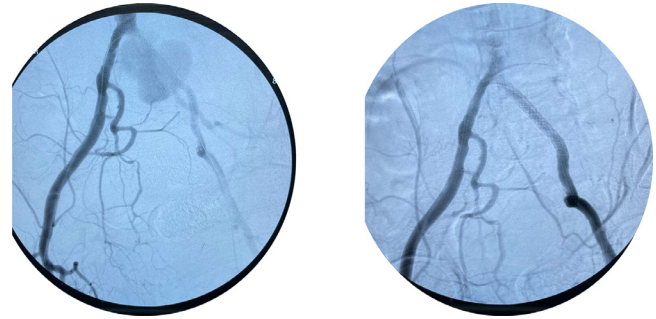


Fig. 2 – Preprocedural aortogram shows a pseudo-aneurysm in the left common iliac artery/aortogram obtained immediately after stent-graft placement, the left common iliac artery aneurysm was completely excluded with coverage of hypogastric artery origin.

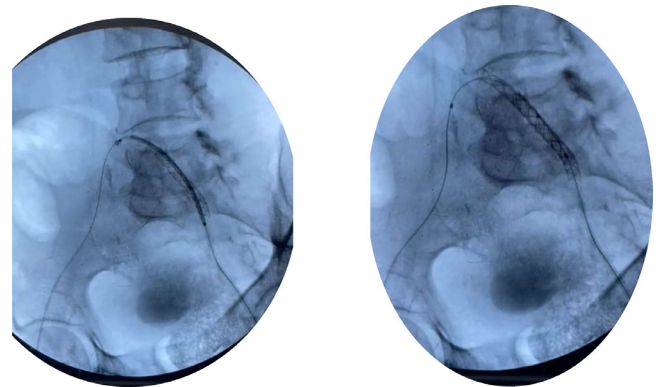


Fig. 3 – Placement of the covered stent in the proximal portion of the stent graft.

covered stent was placed in the proximal portion of the stent graft and post-deployment dilatation was performed with a 7-80 mm balloon catheter intra-stent and in the overlapping zone. The aneurysm was completely excluded as confirmed by on-table angiography; however, the origin of the ipsilateral hypogastric artery was covered with an endo-graft limb. No additional treatment was given.

The patient's general condition, including clinical signs of infection and anemia, gradually improved after the endovascular procedure. Antibiotic therapy was continued.

Four days after the endovascular procedure, the patient presented with septic shock of pulmonary origin and was admitted to intensive care.

The patient was on oral anticoagulation.

A CT scan confirmed that the stent was patent with no evidence of endoleak (Fig. 4).

Computed tomography and ultrasound-guided percutaneous drainage of the psoas abscess was performed, and the patient had a positive culture and the isolate was *C. albicans*.

Blood cultures were negative. However, the urine culture grew *C. albicans*.

The patient was started on antifungal therapy.

After 6 weeks in hospital, the patient died of septic shock.

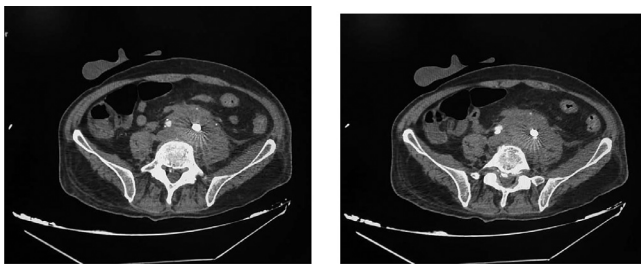


Fig. 4 – Contrast-enhanced CT scan 1 month later shows no evidence of contrast extravasation or endoleak signs.

Discussion

Osler first used the term “mycotic aneurysm” to describe a patient who died of fever, chills and pneumonia, and whose autopsy revealed vegetation on the aortic valve and four aortic arch aneurysms covered with “fresh fungal vegetation”. Today, fungus is rarely the mechanism responsible for an infected aneurysm; however, the term “mycotic aneurysm” is still used and was understood to mean any infection caused by microorganisms [3,4].

Mycotic aneurysms as a result of microbial arteritis are rare, with a reported incidence of 0.06-0.65% [5]. In particular, peripheral artery involvement is much less common than abdominal aorta involvement. In the current era of modern antibiotics, they are rare [6].

Only 2% of mycotic aneurysms have true fungal infections origin [3] and these mainly affect the aorta and cerebral vasculature.

To our knowledge, only 2 other isolated aneurysms of the common iliac artery infected by *C. albicans* have been reported in the literature [7,8]. In Fig. 5, we have tried to group the two observations in the form of a table, showing the age and sex of the patients, their medical history, the additional examinations used to diagnose a *C. albicans* infection, how the aneurysm was managed and the pathophysiology of the infection.

Risk factors for the development of infected aneurysms include arterial trauma from any cause, which accounts for 29% of cases [9], immunosuppression secondary to chronic diseases such as diabetes, malignancy, alcoholism, collagen vascular disease, AIDS and steroid therapy in 24% of cases [9], concurrent sepsis with 17% [9], endocarditis also accounts for 17% [9], congenital cardiovascular defects with 10% [9] and primary 3% [9].

In particular, with regard to systemic fungal infections, examples of multiple foci have been reported in the literature: Fungal endocarditis [10,11], central line infection, colonoscopy polypectomy [12], parental hyperalimentation [7]. and as risk factors for invasive *Candida* infection: immunodeficiency, critical illness and prolonged antibiotic exposure [13].

Intertrigo is a common inflammatory dermatosis of the opposing skin surfaces that can be caused by a variety of infectious agents, particularly *Candida*, under the influence of mechanical and environmental factors. A variety of predisposing factors, particularly obesity, diabetes mellitus and immuno-

suppressive conditions, facilitate both the occurrence and recurrence of disease [14].

Our patient was a high-risk candidate for invasive candidiasis due to diabetes and prolonged corticosteroid use. Four pathophysiological mechanisms of mycotic infection have been proposed [15]:

- Infectious microemboli in the vasa vasorum of an artery of normal caliber or with a pre-existing aneurysm (typical example of infective endocarditis).
- These septic emboli lead to inflammation and necrosis of the media and adventitia, followed by aneurysmal dilatation.
- Infection of the pathological intima (ulcerated atherosclerotic plaque or mural thrombus) by infectious agents circulating in the arterial lumen during bacteremia.
- Arterial infection caused by extension of a contiguous infection (spondylodiscitis abscess, pancreatic abscess, pneumonia, etc.) via lymphatic vessels.
- Direct infectious inoculation through trauma to the arterial wall, after penetrating vascular injury (stab wounds, gunshot wounds), intravenous drug abuse or iatrogenic medical manipulation procedures (arterial catheterization or surgery).

The exact origin of our patient’s aneurysm cannot be determined; there are several theories, but the most likely cause is that systemic candidemia, the consequence of a urinary tract infection, seeded both the iliac artery and the psoas. It is also plausible that the iliac artery infection is caused by extension of a contiguous infection from the left ureter and psoas abscess.

The management of “mycotic” aneurysms should ideally eliminate the source of infection and maintain circulation to the area supplied by the artery [16].

Standard surgical treatment of infected aneurysms consists of ligation, excision and debridement of all infected tissue with creation of extra-anatomical or in situ revascularization with autologous material, synthetic (impregnated or silver) grafts or homograft, followed by long-term antibiotic therapy [17].

Intensive systemic antibiotic therapy is essential for successful treatment and should be initiated perioperatively; a broad-spectrum antibiotic should be used until culture susceptibility reports are available and a specific antibiotic is available determined [18].

The required duration of antibiotic therapy is not well defined; recommendations range from 6-8 weeks to life depending on clinical, radiological and hematological evidence infection [3].

The use of a woven Dacron graft impregnated with antibiotics has been reported to prevent graft infection even during in situ replacement [19–21]. Rifampicin is also commonly used to impregnate prosthetic woven Dacron grafts. It has considerable anti-staphylococcal activity and has been shown to bind significantly to gelatin-sealed Dacron grafts [22]. A cryopreserved homograft is known to be more resistant to bacterial colonization and has better mechanical properties than a prosthesis grafts [23]. A silver-coated polyester prosthesis is also available for the management of arterial infections and

		Case 1	Case 2
Gender		Male	Male
Age		70 yr.	49 yr.
Medical history		Systemic candidemia infected through intravenous hyperalimentation catheter 2 years ago.	Diabetes mellitus. Fungal urinary tract infections history. Recurrent right Knee pain and swelling.
Paraclinical findings	Computed tomography	Right hydronephrosis and right solitary common iliac artery aneurysm.	Right hydronephrosis and a 4.6 cm isolated aneurysm of the right common iliac artery
	Bacteriological exam	Candida albicans grew in the resected aneurysm.	Candida albicans grew in the resected aneurysm and the knee effusion. Urine culture also grew candida albicans.
	Histopathological exam	Abscess formation around the aneurysm with phagocyte infiltration in both outer media of aneurysmal wall and vaso-vasorum.	
Intervention		Common iliac artery ligation and resection of the aneurysm	Resection of the aneurysm and common iliac artery ligation without vascular restauration.
Physiopathological mechanism presumed		Systemic candidemia	Direct infection extension from the right ureter or by hematogenous or lymphatic spread.

Fig. 5 – Table that gathers the two cases reported in the literature of an isolated aneurysm of common iliac artery infected by *Candida albicans*.

favorable results have been reported [24]. The optimal management of infected aneurysms remains controversial.

Open surgery has been considered the conventional strategy for repair, but some studies have shown more complications with open surgery, with an in-hospital mortality rate of 30-40% [25]. With the recent rapid development of endovascular techniques, endovascular exclusion of mycotic aneurysms using covered stents is becoming a minimally invasive alternative to open surgery with favorable short and long-term outcomes results [26].

Although the avoidance of the need for deep pelvic dissection, with the associated risks of visceral, genitourinary and pelvic venous injury, is the potential benefit of the endovascular approach in anatomically appropriate cases patients [27].

Our patient was at high risk for open surgery. We opted for a less invasive endovascular approach; 2 covered stents were successfully placed through a percutaneous approach under local anesthesia.

The use of endoluminal technology has the potential benefit of reducing the existing morbidity and mortality associated with infected aneurysmal disease by avoiding general anesthesia, open repair, significant blood loss, full heparinization and prolonged distal ischemia [28].

Potential problems include lack of debridement of infected tissue, which can lead to infection of the stent graft with possible subsequent fracture above or below the device, and the overall concept of introducing foreign material into an infected field. Migration, durability, stent deformation, gross

oversizing or under sizing, and operator experience are also important factors to be considered [29,30].

Complications during endoluminal repair procedures may result in internal iliac artery occlusion.

Sacrifice of the internal iliac artery has been associated with colon and spinal cord ischemia, hip and buttock claudication, impotence, erectile dysfunction, paraplegia and pelvic necrosis [31–34]. However, because of the extensive collateral network in the pelvis, these complications are relatively rare.

Aneurysms of the common iliac artery extending within 1 cm of the internal iliac artery preclude graft placement proximal to the internal iliac artery orifice. In such cases, the ipsilateral internal iliac artery is occluded with embolization coils to prevent retrograde flow into the aneurysm, leading to aneurysmal expansion and potential rupture, and the graft is extended into the external iliac artery [35].

Coil embolization allows for a more proximal occlusion, preserving the arteriolar and capillary beds, potentially minimizing the risk of lumbar and pelvic ischemia or necrosis [36].

In the case of an internal iliac artery aneurysm, the distal branches must also be occluded and then, in addition to coil placement, direct thrombin injection into the aneurysm may facilitate complete thrombosis [32].

However, the internal iliac artery can also be inadvertently occluded during endoluminal repair procedures due to trauma, embolization or inadvertent endograft coverage [35].

Our patient was not ambulatory due to a long stay in intensive care and therefore never had any symptoms related to the internal iliac artery occlusion.

Conclusion

Mycotic pseudoaneurysm of the iliac arteries is extremely rare, especially of fungal origin. In our case we opted for endovascular exclusion of the aneurysm, which is the optimal option in patients with comorbidities, as open surgery is associated with higher mortality.

Patient consent

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.

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