Case Report
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Tuberculous Aortitis in Immunocompromised Patient

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Abstract

Herein we report a case of an immunodeficient patient with Mycotic Aortic Aneurysm (MAA). A 34 year-old male, recently immigrated from Ivory Coast to Italy, was referred to our hospital for a painful abdominal swelling. Computed-tomography-angiography revealed multiple saccular pseudoaneurysms of the descending thoracic and abdominal aorta and infra-renal aneurysm, complicated with visceral arteries occlusive disease and right common iliac artery thrombosis. The patient underwent combined endovascular aneurysm exclusion and open revascularization of visceral vessels and right iliac artery. MAA is a rare life-threatening disease. Surgical management of MAA with thoracoabdominal involvement remains challenging, with no general consensus on the technique of choice.

Keywords: Mycotic aortic aneurysm; Infra-renal aneurysm; Endovascular aneurysm exclusion; Open visceral revascularization; Thoracoabdominal aneurysm; Tubercular aortitis; Immunodeficiency; Chronic aortic rupture; Tuberculosis; Immunocompromised patient

Abbreviations

MMA: Mycotic Aortic Aneurysm; CMV: Cytomegalo Virus; HBV: Hepatitis B Virus; CTA: Computed-Tomography-Angiography; IGRA: Interferon-Gamma-Release Assay; GSV: Great Saphenous Vein; PTFE: Polytetrafluoroethylene; HAART: Highly Active Antiretroviral Therapy; TAAA: Thoracoabdominal Aortic Aneurysm

Introduction

Mycotic Aortic Aneurysm (MAA) is a rare life-threatening vascular disease, difficult to diagnose and challenging to treat. It has the tendency to grow rapidly and to rupture [1]. A mortality rate of 16% to 44% without surgical treatment has been reported [2]. Hospital mortality may also be significant due to complexity of surgical management and to severe medical comorbidities. In presence of suspicion of infectious etiology, large spectrum antibiotic therapy must be started [3]. The antibiotic administration combined with surgical reconstruction is the management of choice in these cases [3].

Herein we present a case of tuberculous aneurysm in immunocompromised patient.

Case Report

A 34 years old male patient, recently immigrated from Ivory Coast, was referred to our hospital in urgency with a painful abdominal swelling. He reported the swelling to be appeared first two years before, together with a lymphedema of the right leg. He complained of claudication of the same limb. During his stay in the hospital, laboratory findings showed erythrocytes 3.410.000/mm^3, leucocytes 4.580/mm^3 (neutrophils 60%, lymphocytes 32%), hemoglobin 9.8 g/dl, MCV 88 fl, CD4 24%, CD8 38%, CD4/CD8 0.63. Serological test was positive for HIV (Human Immunodeficiency Virus), CMV (Cytomegalo Virus), and HBV (Hepatitis B Virus) infection. Blood culture was positive for Serratia Marcescens. Computed-Tomography-Angiography (CTA) revealed: multiple saccular pseudo-aneurysm in the descending thoracic aorta and in the abdominal aorta; (Figure 1) a 4.6 cm infra-renal aortic aneurysm; diffuse thickening of the aortic wall; celiac artery dissection and superior mesenteric artery occlusion; thrombosis of common and external right iliac arteries; patency of the inferior mesenteric artery; huge thrombosed pseudo-aneurysm adherent to the distal aorta and the right psoas muscle (20x12x8.5 cm); obstruction of superior vena cava and common right iliac vein; osteolytic lesions of L2 and L3 vertebrae. Hypertrophic-dilative cardiomyopathy was documented and further examinations revealed mild heart failure. As soon as the Interferon-Gamma-Release Assay (IGRA) resulted positive, suggesting tuberculosis, anti-tuberculosis therapy was started.

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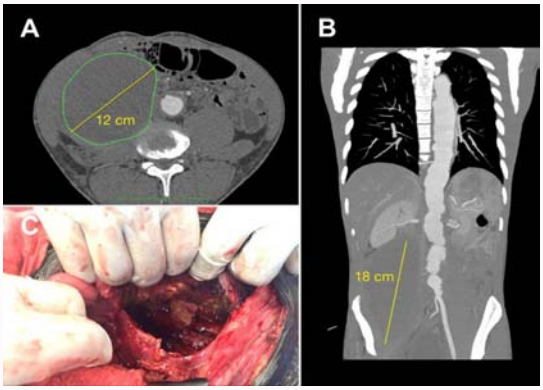

Figure 1: Vast pseudoaneurysm occupying large part of the right abdomen. A: axial view. B: coronal view. The irregular profile of the aorta and the right iliac axis thrombosis are visible. C: pseudoaneurysmatic sac after removal of thrombotic material, accessed through a separate pararectal incision.

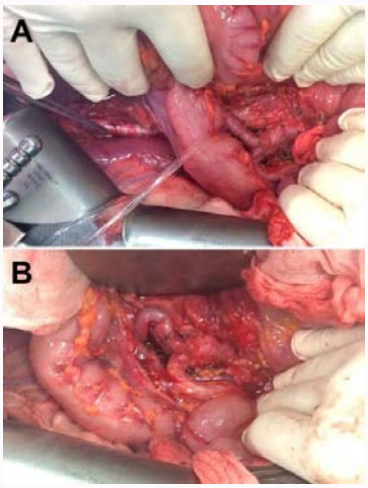

Figure 2: A: exposure of inferior mesenteric artery and left common iliac artery. The vascularization of celiac-mesenteric territory is supplied by the left colic artery, which appears hypertrofic. B: anastomosis on the inferior mesenteric artery of the saphenous vein bypass from left common iliac artery.

Based on the laboratory and imaging findings, we concluded that the thoraco-abdominal aortic disease was probably caused by tubercular dissemination and the abdominal pseudoaneurysmatic mass was consequence of a chronic aortic rupture. The case was discussed in a multidisciplinary meeting and a totally open surgical option was ruled out as it would have entailed total replacement of descending and abdominal aorta, which was too risky for a patient with heart failure and disseminated infection. The visceral vascularization guaranteed only by the inferior mesenteric artery precluded the standard procedure of abdominal aortic aneurysm exclusion with endovascular prosthesis. For these reasons, the patient was treated with hybrid approach.

The procedure was performed in one stage, under general anesthesia with the patient placed in supine position. The abdominal aorta and left common iliac artery were exposed through transperitoneal abdominal approach. Great Saphenous Vein (GSV) was used as graft material for the left iliac artery-to-inferior mesenteric artery bypass (Figure 2). A mobile digital C-arm image intensifier was used. An aorto-uniliac endoprosthesis (Cook Zenith, William A. Cook Australia, Ltd., Brisbane, Australia) was deployed to exclude the infra-renal aortic aneurysm and to seal the aortic breach

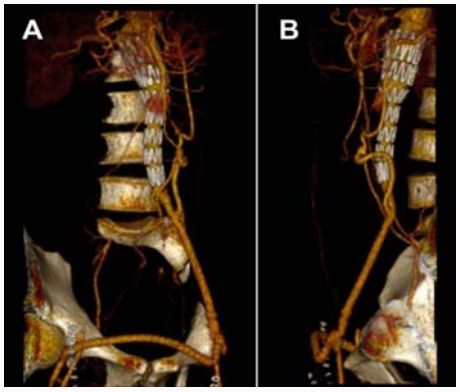

Figure 3: Volume rendering of postoperative CT-A shows exclusion of infrarenal aortic aneurysm with aorto-uniliac endoprosthesis, patency of left iliac artery-to-inferior mesenteric artery bypass and femoro-femoral crossover bypass.

that feeded the pseudoaneurysm. The right leg was revascularized with a femoro-femoral crossover bypass with Polytetrafluoroethylene (PTFE) graft 7 mm. A para-rectal incision has allowed the removal of thrombotic material from the pseudoaneurysmatic sac (Figure 1C). The patient was evaluated with post-procedure CTA that showed a good patency of the bypasses (Figure 3). One week after surgery, the patient was transferred to the infectious disease unit, where he continued antibiotics and antiretroviral therapy. Two months later he was discharged with Highly Active Antiretroviral Therapy (HAART).

Discussion

MAA is a rare life-threatening vascular disease, accounting for less than 1% of all aortic aneurysms [4]. Unspecific symptoms (abdominal or lumbar pain, fever, weight loss) make early diagnosis difficult [5]. The most common pathogens causing MAA are *Staphylococcus aureus*, *Streptococcus* species, *Salmonella*, *Pseudomonas*, *Escherichia Coli* [4]. *Micobacterium tuberculosis* is a rare cause of MAA that can involve the thoracic and abdominal aorta with similar frequency, whereas the involvement of the aortic arch and the ascending aorta is less common [6]. The immunodeficiency of our patient has probably facilitated the tubercular dissemination to the aortic wall [6]. The tubercular process may involve the aorta either by direct extension of pathogens from an adjacent area or through haematogenous spread [7]. The most common cause is the erosion of the aortic wall from the outer side due to a contiguous focus, such as an abscess originating from paraspinal lymph nodes, lung, pericardium, or vertebrae [3]. In our patient, there was an osteolytic tuberculous process of L2-L3 vertebrae likely involving paraspinal lymph nodes. This caused the aortic wall erosion resulting in a massive pseudoaneurysm. The CTA showed that the retroperitoneal mass extended from L2 vertebra to the level of femoral heads, was adherent to the right psoas muscle, and caused the compression of the right iliac vein. Even if the CTA did not show any contrast enhancement of the pseudoaneurysm, based on its size and tenderness we believed that it had been still growing due to blood dripping through an aortic breach. The presence of right leg claudication due to complete thrombosis of the right iliac artery warranted the revascularization through femoro-femoral bypass. The patency and the good calibre of the inferior mesenteric artery allowed the reconstruction of intestine blood supply through left iliac-to-inferior mesenteric artery bypass. The multiple pseudo aneurysmatic ectasia and the infrarenal aortic aneurysm were treated with aorto-uniliac endoprosthesis.

MAA has high mortality (over 55%) without surgical intervention [8]. Classically, its treatment is based on combined open surgery and antibiotic therapy [3]. Antibiotic administration should be started as soon as its infectious etiology is suspected. A blood culture will confirm the presence of bacteremia, and guide the correct antibiotic selection. There is no consensus about the duration of postoperative antibiotic therapy. Patients undergoing endograft placement need lifelong surveillance for the high risk of infection [9]. Open repair of Thoracoabdominal Aortic Aneurysm (TAAA) was the technique of choice in the past decades. Despite the improvement of surgical techniques and organ protection methods, open surgical treatment has still a 40% rate of mortality, because of the extensive surgical exposure, severe medical comorbidity, and unstable patient condition due to sepsis [9]. Surgical options include wide resection of the infected tissues and *in situ* reconstruction with prosthetic graft or extra-anatomic bypass [3]. Nowadays, the endovascular treatment can be used either as a bridge or as a definitive treatment. In the acute setting, the deployment of a stent graft can achieve hemodynamic stability, while definitive surgical treatment is postponed until sepsis is controlled [10]. The endovascular repair is minimally invasive and provides rapid aneurysm exclusion [11]. However, more advanced endovascular solutions are sometimes required, such as branched-fenestrated stent grafts or chimney techniques. These techniques are technically challenging and time-consuming, which increases the risk of aneurysm rupture in urgent cases [12]. Moreover, the deployment of synthetic graft material in an infected site is associated with higher risk of recurrent infection and shorter graft patency [9,11]. To limit this risk, the graft materials can be soaked in antibiotic solution before being implanted.

We had to combine open approach together with endovascular procedure to guarantee the visceral vascularization, since the celiac trunk and the superior mesenteric artery were occluded. This was done through a retrograde bypass from the left iliac artery to the inferior mesenteric artery. In fact, the celiac-mesenteric circulation was mainly supplied by the inferior mesenteric artery. The choice of autologous material was made in order to reduce the risk of infection as the site was potentially contaminated. Published literature on MAA show that hybrid approach is feasible and probably associated with a lower complication rate than totally open or totally endovascular repair [13]. Rosenblum et al., published encouraging results of the hybrid technique [14]. They reported improved short-term outcomes in patients treated with hybrid approach compared with patients treated with traditional surgery. Böckler et al., found that hybrid repair is encouraging for select high-risk patients with complex and extended TAAAs [13]. In our case, the totally open surgical option would have been too risky due to the patient's comorbidities and to the anatomical difficulties to approach the thoracic aorta. In addition, the extensive disease of aortic branches put the patient at high risk of ischemia of abdominal organs, spinal cord, pelvis and lower limbs in case of aortic cross-clamping. By avoiding aortic cross clamping, the hybrid approach permitted to avert ischemic complications as well as the elevated risk of heart failure related to the poor cardiac conditions.

Conclusions

The management of MAA still remains controversial due to the severity of this condition. Each patient should be treated on an individual basis. The reported case required a hybrid approach because of the comorbidities and the anatomical features. Larger case series and a longer follow up are required to increase the knowledge on the durability of aortic stent graft with visceral vessels reconstruction in MAA.

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