Ligation of the aorta for a mycotic abdominal aortic aneurysm in an infant

Zahira Zouizra, Soukaina Benbakh, Gaël Biaou and Drissi Boumzebra

Abstract
Mycotic aortic aneurysms are exceedingly uncommon in infants and they have a high risk of rupture. Their surgical management is extremely challenging. We report a case of a 22-month-old girl who presented with abdominal pain and fever revealing a ruptured mycotic aneurysm of the infrarenal aorta. The surgical treatment consisted of a ligation of the proximal and distal ends of the aneurysm. Postoperative course was significant for hypertension. A year and a half follow-up showed no other complications. Limited data are available concerning our chosen technique, but the reported cases showed a good short-term outcome.

Keywords
Abdominal aorta, mycotic aneurysm, ligation of the aorta, infant aneurysm

Introduction
Abdominal aortic aneurysms (AAAs) are usually diagnosed in the elderly. It is extremely uncommon in infancy and early childhood.1,2

Rarely, pediatric AAAs are congenital. More commonly, they are acquired secondary to an underlying process: infection, inflammation, trauma, iatrogenic and systemic diseases (Takayasu’s arteritis, Kawasaki disease, Behçet’s disease) and connective tissue disorder (Marfan, Loeys–Dietz and Ehlers–Danlos syndromes).

Mycotic aneurysms represent 2.6% of aneurysms in all age groups3 and they are even rarer in the pediatric population. Prior to the development of antibiotics, the causes of mycotic aneurysms were dominated by endocarditis. Nowadays, the most reported cases are secondary to an iatrogenic factor, frequently umbilical catheterization.4

Literature reviews usually consist of solitary case reports or limited series which makes the optimal surgical treatment of AAAs poorly defined. Here, we present the case of a 22-month-old infant with a ruptured mycotic aortic aneurysm which was successfully treated with surgical ligation of the aorta.

Case report
A 21-month-old girl presented to a secondary care hospital with an isolated fever that was treated as a bacterial pulmonary infection. She was discharged and put on a long-term course of oral antibiotics after she was afebrile for 2 days.

A month later, she presented to the health facility with worsening abdominal pain and fever. There was no history of nausea, vomiting or any other associated sign. Abdominal ultrasound and angio-computed tomography (CT) revealed an AAA measuring 40 mm in diameter (Figure 1). She was referred to our hospital for surgical management of the aneurysm.

On admission, the patient presented with hemodynamic shock. She was pale, tachycardiac and hypotensive. She was admitted immediately to the intensive care unit. An emergency CT scan revealed a ruptured infrarenal aortic aneurysm with a giant retroperitoneal hematoma measuring $93 \times 54 \times 92 \text{ mm}^3$. She underwent urgent surgery after hemodynamic stabilization.

Surgical exploration found a giant AAA compressing on approximate organs with signs of inflammation and rupture.
The aneurysm was dissected firsthand and then the aorta was ligated below the origin of the renal arteries and above the iliac bifurcation with 5-0 silk after verifying the collateral circulation on the angio-CT. The remaining aortic aneurysm sac was resected. The abdominal cavity was rinsed and closed after verifying the local hemostasis (Figure 3).

The patient was put on intravenous (IV) antibiotics: a combination of amoxicillin, clavulanic acid and gentamicin. Results of the laboratory tests showed anemia with hemoglobin at 10.5 mg/dL, white blood cell count at 30,000 cells/µL, erythrocyte sedimentation rate at 52 mm/hh and C-reactive protein at 142.78 mg/L. Electrocardiography (EKG) was normal and echocardiography showed no signs of endocarditis.

A postoperative angio-CT showed a persistent retroperitoneal hematoma measuring $70 \times 50 \text{mm}^2$. Clinical assessment showed no signs of lower limb ischemia. The patient’s postoperative course was remarkable for hypertension which was treated with the combination of beta-blockers, angiotensin-converting enzyme (ACE) inhibitors and calcium channel blockers (CCBs). No other complications were noted.

The patient was discharged home on postoperative day 18. She was put on pain medication and beta-blockers for 3 months thereafter.
The pathologic examination of the aortic tissue showed inflammatory signs with suppuration and made a conclusion of the mycotic aneurysm. Microbiological cultures of the aortic tissue and blood cultures were both sterile.

Discussion

In 1885, Sir William Osler for the first time introduced the term “mycotic aneurysm” describing a case of multiple aneurysms resulting from a “malignant endocarditis.”

A classification of childhood arterial aneurysms was elaborated by Sarkar based on a review of 135 reported cases of aneurysms with a variety of localizations and causes. He regrouped the aneurysms caused by arterial infection into class I. He also described them as having a higher potential to expand and a greater risk of rupture due to the weakening of the arterial wall by the infectious process.

Mycotic aortic aneurysms can result from a systemic infection such as endocarditis and bacteremia or they can be secondary to a perivascular infection such as an adjacent abscess or an iatrogenic source of infection such as umbilical catheterization. Coarctation of the aorta predisposes an individual to mycotic aneurysms, especially when associated with a bicuspid aortic valve.

Mycotic AAAs can be symptomatic and present with fever which may or may not be associated to abdominal pain, hypertension, the presence of an abdominal mass, sepsis or shock subsequent to rupture. However, it can also be discovered incidentally on a physical examination or abdominal ultrasound.

Staphylococcus and streptococcus bacteria have been reported as the most frequent infective organisms. In some cases, such as ours, the bacterial agent causing aneurysm is unidentified because of a prior antibiotic course.

There are multiple approaches described for treating AAAs in children: endovascular approach with stent grafting, open repair with the use of a prosthetic material, a cryopreserved human allograft, an aortoplasty with a patch or simple ligation of the distal and proximal ends of the aneurysm. They are all not suitable for treating mycotic aneurysms, which make it a case-by-case decision in the absence of recommendations and treatment algorithms.

Open repair with interposition of a GORE-TEX or polytetrafluoroethylene (PTFE) graft and reconstruction with a cryopreserved human allograft are the most used methods. However, the risk of infection makes them impractical in mycotic aneurysms.

Ligation of the ends of the aneurysm and extra-anatomic bypass grafting can be used in the case of mycotic aneurysms to permit revascularization in a sterile field, but the graft
cannot assure a sufficient blood flow as adulthood is reached and a second surgery may be required.10

The isolated ligature of the ends of the aneurysm can be performed without short-term complications in infrarenal aortic aneurysms. A later construction can be necessary.10

Limited data are available on the long-term results. Park published the first case of aortic ligation as the surgical treatment of a ruptured mycotic aneurysm in a 9-year-old girl in 1981. No short-term complication was noted but it was not reported if an additional surgical intervention was required.5

Our case was characterized by the urgency of the surgical treatment. It consisted of debridement of the aneurysm and ligature of its proximal and distal orifices. The following factors contributed to this decision:

1. The known risk of graft infection, especially in emergency surgery, without prior infection control;
2. A graft repair in a 22-month-old child will not have the ability to expand while she grows up;
3. The unavailability of cryopreserved human aortic allografts in our center.

A year and a half of postoperative follow-up was uneventful and was without signs of lower limb ischemia, exercise-induced fatigue or growth retardation. A long-term follow-up is necessary and a secondary reconstruction may be required.

Conclusion

In conclusion, mycotic aortic aneurysms in infants are rare and have a higher potential for rupture. They are mostly presented in the literature as isolated cases or short series making the elaboration of recommendations hard. The surgical treatment of mycotic aortic aneurysms is well defined in the adult population, but many approaches are not suitable in children.

Our case was particularly challenging because of the urgency of the surgery not giving us enough time to control the infection. We adopted a simple approach with obvious good short-term results. Only a long-term follow-up can provide a proper assessment of this technique.

Declaration of conflicting interests

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Ethical approval

The authors declare that our institution does not require ethical approval for reporting individual cases.

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Informed consent

The authors declare that written informed consent was obtained from a legally authorized representative for anonymized patient information to be published in this article.

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