Case Report

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Successful surgical management of coarctation of the aorta with infective endaortitis and splenic abscess: a case report

Xuebin Wang[1], Yongjun Li[2,*] and Yuexin Chen[1,*]

Abstract

Infective endaortitis with a splenic abscess is an extremely rare and fatal complication of coarctation of the aorta. We herein describe a 19-year-old female patient with a 2-month history of intermittent fever with progressive abdominal and leg pain. Aortic computed tomography angiography showed a rapidly progressive poststenotic saccular aneurysm immediately distal to the coarctation. Enhanced abdominal computed tomography showed a severe splenic abscess. During the operation, the infected spleen was resected first. We subsequently removed all of the diseased aorta and performed in situ aortic reconstruction with an artificial tube graft and without extracorporeal circulation. The patient had an uneventful postoperative clinical course and recovered very well during the 4-year follow-up. In summary, our case report details the successful surgical treatment of coarctation of the aorta with infective endaortitis and a splenic abscess and thoroughly discusses the indications and key notes for surgery without extracorporeal circulation in this complex disease.

Keywords

Coarctation of the aorta, infective endaortitis, splenic abscess, surgical treatment, extracorporeal circulation, computed tomography angiography

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[1]Department of Vascular Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing, China
[2]Department of Vascular Surgery, Beijing Hospital, National Center of Gerontology, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing, China

*These authors contributed equally to this work.

Corresponding author:
Yuexin Chen, Department of Vascular Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, #1 Shuai-Fu-Yuan, Wang-Fu-Jing, Beijing 100730, China.
Email: cyuexin2007@163.com

Introduction

Congenital coarctation of the aorta (CoA) accounts for 4% to 5% of all congenital heart diseases.[1] CoA tends to be complicated by hypertension, heart failure, ascending aortic aneurysmal dilatation, and rarely endaortitis.[2] Infective endaortitis with a splenic abscess is an extremely rare and fatal complication of CoA. Few reports have described this complication.[3] We herein report a case involving a 19-year-old girl who was successfully cured of this complex disease.

Case report

A 19-year-old female patient with an unremarkable medical history was referred to the vascular surgery department of Peking Union Medical College Hospital because of a 2-month history of an intermittent fever with progressive abdominal pain and leg pain. At admission, the patient's temperature was normal, and her blood pressure was higher in the upper limb than in the lower limb (160/85 vs. 105/80 mmHg, respectively). An atypical systolic murmur could be heard most strongly over the left subclavian area and the left scapular region. Scattered petechial hemorrhages could be seen on the bilateral lower extremities, especially on the bottom of the feet (Figure 1 (a)). The white blood cell count and high-sensitivity C-reactive protein concentration were 14.1×10^9/L (reference range, 4.0–10.0×10^9/L) and 105 mg/L (reference range, 0–20 mg/L), respectively. The hemoglobin concentration was 83 g/L (reference range, 110–160 g/L). The patient's echocardiographic findings were normal with no evidence of infective endocarditis. Notably, the diagnosis of CoA with a poststenotic saccular aneurysm of the aorta had been confirmed by aortic computed tomography angiography (CTA) in an external hospital 1.5 months before the current presentation (Figure 2(a)), and a splenic infarction was also found.

Treatment was initiated with broad-spectrum intravenous antibiotics. However, on the fourth day of admission, the patient developed a sudden-onset high fever (102.6°F), abdominal pain, and vomiting. Blood analysis revealed a markedly elevated leukocyte count (29.1×10^9/L; reference range, 4.0–10.0×10^9/L) and a high procalcitonin concentration (12.25 ng/mL; reference range, <0.5 ng/mL). A blood culture showed growth of *Viridans streptococci*. A new aortic CTA examination in our hospital showed that the poststenotic saccular aortic aneurysm had substantially grown in size (Figure 2(b)). Additionally, enhanced abdominal CT showed a severe splenic abscess (Figure 1(b)). Based on the patient's clinical manifestations, laboratory results, and imaging evidence, a preliminary diagnosis of infective endaortitis was established. Considering the rapid enlargement of the poststenotic saccular aortic aneurysm, an emergency surgery was scheduled.

During the operation, the infected spleen was resected first. Next, a left posterolateral thoracotomy was performed through the fourth rib bed. The distal aortic arch and the descending aorta were carefully dissociated. Aortic coarctation with a poststenotic saccular aortic aneurysm was confirmed. The adventitia of the aortic aneurysm remained intact, and no obvious infective tissue was present around the aneurysm. After transverse clamping of the distal aortic arch above the constricted segment and the distending aorta distal to the aneurysm, a strong pulse was felt on the distal thoracic aorta, with an acceptable systolic blood pressure of 100 mmHg invasively measured through the dorsal pedal artery. This indicated that more extensive collateral vessels secondary to long-term aortic coarctation could maintain sufficient infusion of the distal aorta even after clamping, as we predicted before the surgery.

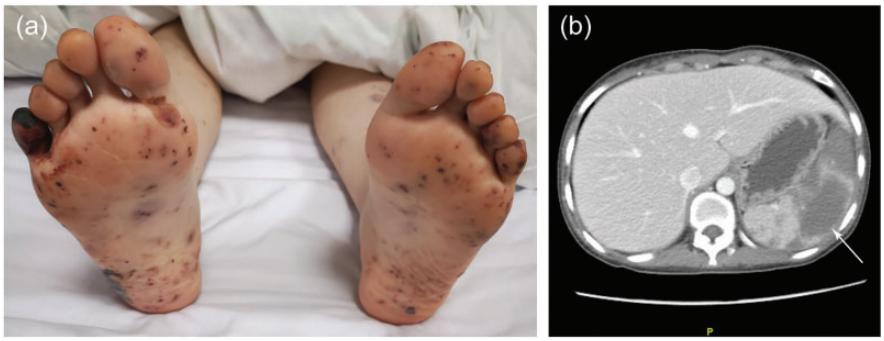

Figure 1. Septic embolization. (a) Scattered petechial hemorrhages were seen on the bottom of the foot. (b) Enhanced abdominal computed tomography revealed a severe splenic abscess (white arrow).

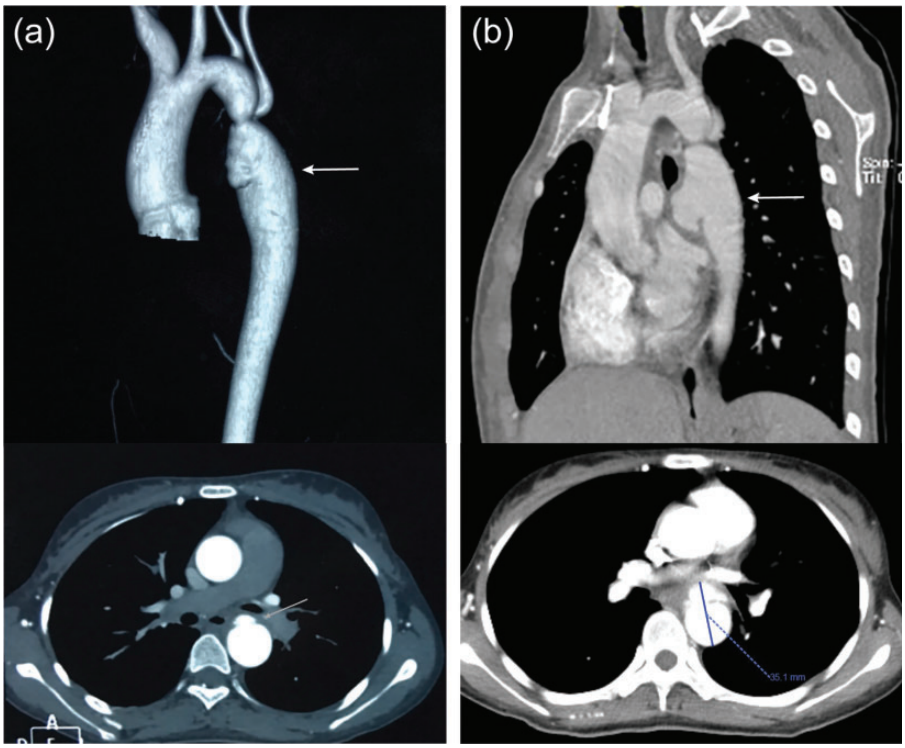

Figure 2. (a, b) Aortic computed tomography angiography showed a noticeably increased size of the poststenotic saccular aortic aneurysm (the maximum diameter of the diseased aorta increased from 29.3 to 35.1 mm) within just 1.5 months (white arrows).

A surgical repair strategy without extracorporeal circulation was conducted. Carefully maintaining an invasive systolic blood pressure ranging from 100 to 110 mmHg as measured through the dorsal pedal artery, which was in accordance with the patient's preoperative ankle blood pressure, we removed all of the diseased aorta and then

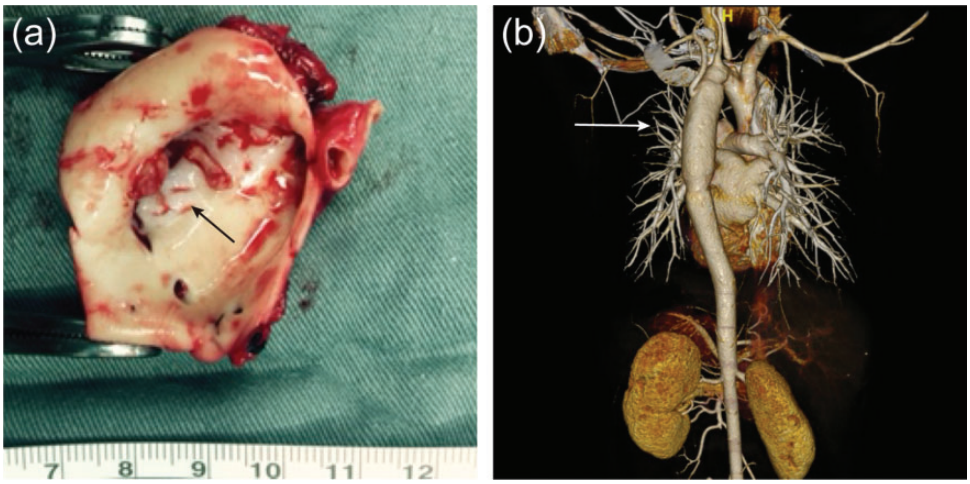

Figure 3. (a) Vegetations and a saccular aneurysm were seen immediately distal to the coarcted segment (black arrow). (b) Aortic computed tomography angiography 4 years later showed an artificial vessel with unobstructed thoracic aortic blood flow (white arrow).

performed in situ reconstruction of the aorta with a Dacron artificial tube graft (20-mm diameter and 40-mm long). The clamping delay was 29 minutes. Macroscopic examination of the operative specimen revealed coarctation and poststenotic saccular aortic aneurysm with several vegetations adhered to its intima (Figure 3 (a)). The postoperative clinical course was uneventful, and intravenous antibiotics were administered for 6 weeks after confirming a negative blood culture. Four years later, this patient was followed up without any events. Aortic CTA demonstrated that the artificial vessel functioned normally (Figure 3(b)). This case report did not require ethics committee approval because it did not involve animal or human studies. Verbal consent was obtained from the patient for publication of this manuscript and any accompanying images and medical data.

Discussion

Infective endaortitis secondary to CoA is extremely rare. The mechanism of CoA causing infective endaortitis is similar to that of infective endocarditis.[4] Infective endaortitis usually occurs in the aortic wall below the adjacent constricted segment. Local blood turbulence causes endothelial injury, which in turn creates conditions for infective endaortitis. Once local infection has occurred, it can lead to local pyogenesis, mycotic aneurysm formation, and sometimes even distal embolization.

No randomized controlled studies have been performed to guide the management of infective endaortitis. Most authors agree that antibiotic therapy in combination with complete surgical excision of the infected aorta is the best treatment choice.[5] Although some good results have been reported with endovascular stent grafts in the management of infective endaortitis,[6,7] there is not yet enough evidence to consider it an alternative to surgery because of the risk of graft infection. Congenital CoA with infective endaortitis is even rarer. This disease combination has only been reported sporadically in the past, and there are no clear guidelines for its treatment.

Complete surgical excision of the coarctation and infected aorta with in situ reconstruction was the most frequently selected and most effective surgical procedure.[2,4,8] Some surgeons also chose to perform extra-anatomic reconstruction, which also achieved good results.[9] However, endovascular techniques were rarely used for this complex pathological condition. The two main concerns were that CoA and mycotic aneurysm needed to be treated at the same time and the infectious lesions could not be completely removed. In the present case, we did not perform extra-anatomic reconstruction mainly because the greater trauma and hemodynamic changes associated with this procedure were unsuitable for our young patient. Additionally, the intact adventitia indicated that in situ reconstruction would be possible. Each case needs to be considered individually and will likely be influenced by the skill set of the surgeon.

Several graft materials are available for aortic repair in the setting of aortic infective disease, including autogenous vein grafts, cryopreserved arterial allografts, rifampicin-impregnated grafts, and silver-coated grafts, among others.[10,11] Two single-center clinical studies in recent years have confirmed that in situ aortic reconstruction with cryopreserved allografts is a viable treatment modality with relatively low morbidity and mortality in patients with native and prosthetic aortic infections.[12,13] However, one study showed poor outcomes, with 28% mortality at 1 month and nearly 40% mortality at 1 year.[14] In the present case, because of the lack of an allograft supply in our country, we chose a Dacron tube to reconstruct the aorta, considering the intact aortic adventitia. Ultimately, we achieved a very good prognosis. Thus, when an allograft is lacking, an artificial graft should be considered an alternative in situ reconstructive material for aortic infective endaortitis.

After transverse clamping of the aorta, extensive collateral vessels secondary to long-term aortic coarctation maintained sufficient infusion of the distal aorta in this case. Therefore, we did not choose to establish extracorporeal circulation during the operation. In addition, the blood pressure of the dorsal pedal artery was carefully maintained, which along with maintenance of the blood pressure of the distal aorta ensured an adequate blood supply to the visceral organs, spinal cord, and lower extremities to prevent severe ischemic events. Preoperative measurements of the blood pressure of both the upper and lower limbs each day helped us to ensure that the optimal range was maintained during clamping to decrease the risk of visceral and medullary ischemia. Shortening the duration of aortic clamping as much as possible may also help to prevent severe visceral and medullary ischemia. O'Brien and Marshall[15] reviewed the surgical treatment of CoA and stated that "the procedure is done through an incision in the left side of the chest and does not require the heart-lung machine." After a literature review of the treatment of CoA in patients with aortic infective endaortitis, we found that most surgeons established an intraoperative left heart bypass or cardiopulmonary bypass to prevent postoperative visceral and medullary ischemia;[2,4,16–19] only a few case reports did not mention the application of left heart bypass or cardiopulmonary bypass.[8,20] Therefore, whether establishment of extracorporeal circulation is necessary during surgery for this complex disease remains controversial and should be considered on an individual-patient basis; it mainly depends on whether the collateral arteries can maintain the blood supply of the distal aorta after clamping. The key factors in a successful procedure are preoperative measurements and prediction, reevaluation after clamping,

and careful maintenance of optimal blood pressure during clamping.

Our patient also had a severe splenic abscess, which were likely caused by the shedding of bacterial vegetations. Considering that the infected spleen would probably lead to bacteremia and even post-transplant graft reinfection, we decided to perform a splenectomy in this patient. Thus, the timing of spleen removal was also an important issue that we needed to consider. We finally decided to treat the splenic abscess by performing a splenectomy on the same day as the aortic replacement for three reasons. First, the patient's aneurysm had progressed rapidly and may have ruptured at any time, and the aortic surgery needed to be completed as soon as possible. Second, the abscess was still confined to the spleen. Third, a further operation on the same day as the aortic replacement prevented the patient from sustaining surgical trauma in two separate procedures.

Conclusion

CoA complicated by distal infective endaortitis is rare. This is the first report to describe in detail the surgical experience of treating CoA with infective endaortitis and a splenic abscess, thus providing a useful therapeutic reference for this rare and complex pathological condition. Although aortic resection and in situ replacement by simple clamping without extracorporeal circulation is not the usual method, it was safe and useful in this case because of the sufficient blood supply of the distal aorta by abundant collaterals. As we stated above, preoperative measurements and prediction, reevaluation after clamping, and careful maintenance of optimal blood pressure during clamping were the key factors in achieving a good result for this patient.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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ORCID iD

Yuexin Chen https://orcid.org/0000-0001-9564-5394

References

1. Forbes TJ, Kim DW, Du W, et al. Comparison of surgical, stent, and balloon angioplasty treatment of native coarctation of the aorta: an observational study by the CCISC (Congenital Cardiovascular Interventional Study Consortium). *J Am Coll Cardiol* 2011; 58: 2664–2674.
2. Idir M, Denisi R, Parrens M, et al. Endarteritis and false aneurysm complicating aortic coarctation. *Ann Thorac Surg* 2000; 70: 966–968; discussion 8–9.
3. Barth H, Moosdorf R, Bauer J, et al. Mycotic pseudoaneurysm of the aorta in children. *Pediatr Cardiol* 2000; 21: 263–266.
4. Owens CG, Johnston N, Campalani GF, et al. Aortic coarctation endarteritis resulting in mycotic pseudoaneurysm and acute mediastinitis. *Acta Cardiol* 2010; 65: 463–465.
5. Lopes RJ, Almeida J, Dias PJ, et al. Infectious thoracic aortitis: a literature review. *Clin Cardiol* 2009; 32: 488–490.
6. Kritpracha B, Premprabha D, Sungsiri J, et al. Endovascular therapy for infected aortic aneurysms. *J Vasc Surg* 2011; 54: 1259–1265; discussion 65.
7. Escobar GA, Eliason JL, Hurie J, et al. Rifampin soaking dacron-based endografts for implantation in infected aortic aneurysms–new application of a time-tested principle. *Ann Vasc Surg* 2014; 28: 744–748.
8. Jaleleddine Z, Sana C, Faker G, et al. Infective endarteritis and false mycotic

aneurysm complicating aortic coarctation. *Ann Pediatr Cardiol* 2012; 5: 197–199.
9. Prifti E, Kuci S, Krakulli K, et al. Pseudoaneurysm of the descending aorta complicating an untreated aortic coarctation. *Ann Thorac Surg* 2015; 99: e3–e5.
10. Clagett GP, Valentine RJ and Hagino RT. Autogenous aortoiliac/femoral reconstruction from superficial femoral-popliteal veins: feasibility and durability. *J Vasc Surg* 1997; 25: 255–266; discussion 67–70.
11. Vogt PR, Brunner-La Rocca HP, Carrel T, et al. Cryopreserved arterial allografts in the treatment of major vascular infection: a comparison with conventional surgical techniques. *J Thorac Cardiovasc Surg* 1998; 116: 965–972.
12. Ben Ahmed S, Louvancourt A, Daniel G, et al. Cryopreserved arterial allografts for in situ reconstruction of abdominal aortic native or secondary graft infection. *J Vasc Surg* 2018; 67: 468–477.
13. Masabni K, Weaver MR, Kandagatla P, et al. Cryopreserved allograft in the management of native and prosthetic aortic infections. *Ann Vasc Surg* 2019; 56: 1–10.
14. Touma J, Cochennec F, Parisot J, et al. In situ reconstruction in native and prosthetic aortic infections using cryopreserved arterial allografts. *Eur J Vasc Endovasc Surg* 2014; 48: 292–299.
15. O'Brien P and Marshall AC. Coarctation of the aorta. *Circulation* 2015; 131: e363–e365.
16. Mosca RS, Kulik TJ, Marshall K, et al. Mycotic pseudoaneurysm associated with aortic coarctation. *J Cardiovasc Magn Reson* 2000; 2: 209–212.
17. Haas B, Wilt HG, Carlson KM, et al. Streptococcus pneumoniae causing mycotic aneurysm in a pediatric patient with coarctation of the aorta. *Congenit Heart Dis* 2012; 7: 71–75.
18. Restrepo MS, Turek JW, Reinking B, et al. Mycotic aneurysm in a child with history of coarctation of the aorta repair. *Ann Pediatr Cardiol* 2014; 7: 138–141.
19. LeNoir B, DeNino WF, Bradley SM, et al. Mycotic aortic aneurysm in a child with aortic coarctation. *J Thorac Cardiovasc Surg* 2015; 149: e44–e46.
20. Anderson AM, Cabell CH and Sexton DJ. Aortic coarctation endarteritis in an adult: case report with cardiovascular magnetic resonance imaging findings and review of the literature. *Clin Infect Dis* 2005; 40: e28–e31.