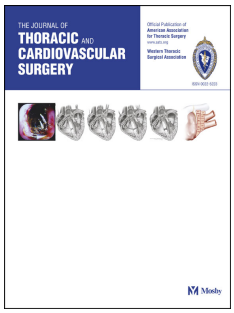


An approach to mycotic aneurysm in unrepaired coarctation of the aorta

Lauren E. Parker, BS, Lillian Kang, MD, Cathlyn K. Medina, BA, Bailey Su, MD, Stephen Miller, MD, Joseph W. Turek, MD PhD MBA, Douglas M. Overbey, MD, Ziv Beckerman, MD

PII: S0022-5223(23)00435-X

DOI: https://doi.org/10.1016/j.jtcvs.2023.05.011

Reference: YMTC 19062

To appear in: *The Journal of Thoracic and Cardiovascular Surgery*

Received Date: 13 March 2023

Revised Date: 30 April 2023

Accepted Date: 3 May 2023

Please cite this article as: Parker LE, Kang L, Medina CK, Su B, Miller S, Turek JW, Overbey DM, Beckerman Z, An approach to mycotic aneurysm in unrepaired coarctation of the aorta, *The Journal of Thoracic and Cardiovascular Surgery* (2023), doi: https://doi.org/10.1016/j.jtcvs.2023.05.011.

Coarctation of the Aorta with Distal Mycotic Aneurysm

Questions to Consider – Set #1

1. What other imaging would you request?
2. When would you repair the pseudoaneurysm?
3. What is the extent of proximal/distal involvement of the pseudoaneurysm and coarctation?
4. What approach would you use to resect the coarctation and pseudoaneurysm?
5. Would you use bypass or standby? Cooling?
6. Would you patch the defect? If so, with what material?
7. How would you achieve a tension free anastomosis?
8. How long would you continue antibiotic therapy?

- Uneventful repair via sternotomy with lower body circulatory arrest
- Pseudoaneurysm recurrence during the same hospital admission

Questions to Consider – Set #2

1. Would you discharge this patient home with small recurrent pseudoaneurysm?
2. What follow-up arrangements would you make?
3. What would be indications for reoperation?
4. What approach would you use, and how would you perform the operation?

An approach to mycotic aneurysm in unrepaired coarctation of the aorta

Lauren E. Parker BS[1], Lillian Kang MD[1], Cathlyn K Medina BA[1], Bailey Su MD[1], Stephen Miller MD[2], Joseph W Turek MD PhD MBA[1], Douglas M Overbey MD[1], Ziv Beckerman MD[1]

[1]Division of Cardiovascular and Thoracic Surgery, Department of Surgery, Duke University Medical Center, Durham, NC

[2]Division of Pediatric Cardiology, Department of Pediatrics, Duke University Medical Center, Durham NC

Address for Correspondence:

Lauren Parker, BS

Duke University Medical Center

2301 Erwin Road, DUMC 3474

Durham, NC 27710

Tel: 1-804-332-4723

Email: lauren.parker@duke.edu

Word Count: 1,000

Acknowledgements: We sincerely thank Megan Llewellyn for providing case illustrations.

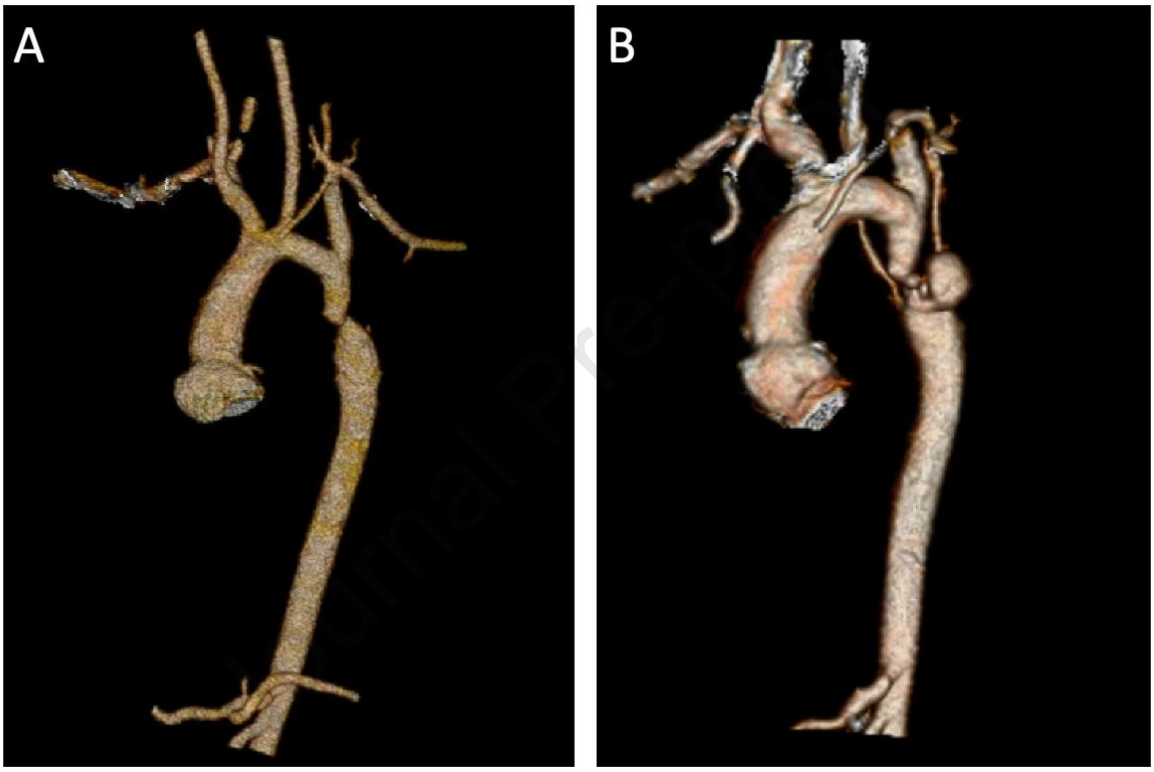
Disclosures: No disclosures to report.

IRB Statement: This case report is approved under Pro00101549 (approved 1/3/2019) with waiver of individual patient consent.

Central Message

Coarctation of the aorta presenting concurrently with mycotic pseudoaneurysm is a rare clinical entity, and surgical management requires careful consideration of timing and operative approach.

Central Picture



Central Picture Legend: Undiagnosed aortic coarctation in a 4-year-old (A) and rapidly developed pseudoaneurysm (B).

Key Words: mycotic pseudoaneurysm, coarctation of the aorta, aortoplasty

Abbreviations: BSA, body surface area; CTA, computed tomography angiogram; TEE, transesophageal echocardiogram

Case Description

This case report is approved under Pro00101549 (approved 1/3/2019) with waiver of individual patient consent. A 4-year-old boy (21.7kg, BSA 0.84m^2) with a history of autism presented to his primary care provider with two weeks of daily fevers. A new, II/VI systolic murmur was auscultated at the right upper sternal border; the patient was otherwise well-appearing. Initial laboratory tests revealed elevated inflammatory markers. Upon these findings, the patient was referred to our emergency department.

Upon arrival, the patient was afebrile and hemodynamically stable. Based on prolonged fever of unknown origin coinciding with new systolic murmur, the patient was admitted to the general pediatrics unit for further work-up. Transthoracic echocardiography identified a bicuspid aortic valve, aortic root dilation to 2.65cm at the sinus of Valsalva, and severe coarctation to 0.4cm at the aortic isthmus with a peak gradient of 90mmHg. There was normal biventricular size/function, and no valvular defects were identified. A chest CT angiogram (CTA) confirmed post-ductal coarctation distal to the origin of the left subclavian artery with prominent collateral vasculature (Figure 1A). Blood cultures were positive for streptococcus pneumoniae, and viral respiratory panel was positive for parainfluenza virus. The patient was started on IV antibiotics.

On hospital day 6, a repeat CTA, cardiac MRI, and transesophageal echocardiogram (TEE) were performed to further evaluate endocarditis versus endarteritis as a source of the patient's bacteremia. On CTA, a new 1.7x1.3x1.5cm area of homogenous contrast enhancement immediately distal to the coarctation was identified, consistent with pseudoaneurysm (Figure 1B). On TEE, a new 3mmx2.4mm vegetation was visualized along the distal aspect of the

coarctation with motion between the post-coarctation aortic lumen and the pseudoaneurysm. The congenital cardiothoracic surgery team was consulted for intervention.

On hospital day 7, the patient was taken to the operating room for coarctation repair and resection of mycotic aortic pseudoaneurysm via median sternotomy (Figure 2A). Cardiopulmonary bypass was established via right atrial and aortic cannulation. The patient was cooled to 18^{o} C, and antegrade cardioplegia was administered into the aortic root with left ventricular vent placement through the right upper pulmonary vein. Dissection of the descending aorta was difficult, as it was immobile due to inflammation associated with the ongoing infectious process. As the transverse aorta was of good diameter, antegrade cerebral perfusion (to both carotid arteries) was achieved by clamping the transverse arch distal to the left carotid artery. The transverse aortic arch across the area of coarctation was opened with longitudinal aortotomy. The pseudoaneurysm neck was identified along the posterior aspect of the aorta immediately distal to the coarctation. Upon exposure, torrential blood flow from collateral vessels was present and returned to the bypass circuit. Probing of the pseudoaneurysm revealed a collection of pus and thrombus. Within the pseudoaneurysm, a 3.0mm tear in the posterior left lateral segment of the descending aorta was visualized (Figure 2B). After debriding as much grossly infected tissue as possible back to viable aortic wall, the tear in the posterior wall of the aorta was repaired with interrupted 6-0 Prolene sutures. A decellularized pulmonary homograft patch was then used to enlarge and repair the aortic coarctation. After irrigating with vancomycin solution, the sternum was reapproximated and the incision was closed in standard fashion. The debrided tissue was sent for culture, which resulted in no growth after 5 days.

The patient progressed as expected in the immediate post-operative period. On postoperative day 10, repeat chest CTA demonstrated pseudoaneurysm recurrence measuring 1.5x0.9x1.6cm. Given the likelihood that pseudoaneurysm recurrence was due to incomplete resection of infected tissue, we elected to continue antibiotic therapy and ensure infection eradication rather than pursue immediate reoperation. The patient remained afebrile and clinically stable. CTA was repeated on post-operative day 18, which demonstrated pseudoaneurysm progression to 2.3x1.2x2.2cm with a neck width of 8mm. Given rapid progression of the pseudoaneurysm in spite of antibiotic therapy, we decided to proceed with reoperation.

A left thoracotomy was chosen for the takeback, and right radial and femoral arterial lines were placed. The chest was accessed through the fourth intercostal space, revealing the recurrent pseudoaneurysm (Figure 3A). The aortic arch and descending aorta were carefully dissected from surrounding adhesions. Partial left heart cardiopulmonary bypass was initiated via descending aorta and left atrial appendage cannulation in order to scavenge blood flow from the robust collateral circulation we encountered in the first operation. The patient was cooled to 34^{o}C, and the proximal descending and mid-descending aorta were controlled with vascular clamps. The entirety of the pseudoaneurysm sac was dissected out from the surrounding tissue and resected from the aortic wall. The resulting aortic defect was repaired with an aortic homograft patch via simple running 6-0 Prolene (Figure 3B). After 22 minutes cross-clamp time, the patient was rewarmed and weaned from cardiopulmonary bypass with appropriate hemodynamics. Repeat CT scanning on POD #9 showed good surgical result with no recurrence, after which the patient was discharged to home on 50mg/kg ceftriaxone twice daily. Outpatient

follow-up with infectious disease and pediatric cardiology included repeat CTA 4 weeks post-op which demonstrated no pseudoaneurysm recurrence, with plans to repeat CTA in 3 months.

Comment

Coarctation of the aorta presenting concurrently with mycotic pseudoaneurysm is a rare clinical entity. While 35 patients have been reported in the literature with a similar presentation, only three were younger than 5-years-old with undiagnosed aortic coarctation and post-ductal mycotic aneurysm, similar to our patient.[1-3] Bacterial infections predominate, primarily *Staphylococcus* and Streptococcus species, however *Mycobacterial* and even *Candida* infections are possible, albeit more rare. Successful surgical treatment of infected vasculature requires wide debridement of all involved tissue, and careful consideration of approach to optimize access to infected tissue (Figure 4).[4] First, deciding when to perform surgical intervention is critical. While we would typically wait until blood cultures became negative, the rapid progression of this patient's aortic pseudoaneurysm prompted urgent surgical intervention. Recommendations for antibiotic duration in more stable mycotic pseudoaneurysms range from 6 weeks-6 months, with some patients requiring lifelong antibiotic therapy.[4]

Surgical approaches to aortic coarctation vary, including median sternotomy, posterolateral left thoracotomy, and clamshell incisions, each with potential pros and cons. We elected to approach the aorta via median sternotomy due to the robust experience we have in this technique for arch reconstruction in neonates and young children. It provides the ability to safely initiate bypass, perfuse the brain via antegrade cerebral perfusion, and salvage blood to the pump. That said, in this case, the difficulty in mobilizing the descending aorta due to the severity of the

inflammatory/infectious process made controlling the descending aorta challenging, ultimately requiring lower body circulatory arrest. Posterolateral left thoracotomy can provide excellent distal visualization of the coarctation and associated pseudoaneurysm; our initial concern was that upon entering the chest, we would encounter unexpected bleeding from the pseudoaneurysm, with no ability to control the proximal aorta and quickly go on bypass. A clamshell incision offers excellent visualization and mobilization and is often used for adults in arch/descending aorta replacement, but the main downside is morbidity associated with the incision itself.[5] Finally, though endovascular stenting has been used in children with mycotic aneurysm, this approach was deemed inappropriate in the setting of an active infection.[6, 7]

Another important consideration is what material should be used for surgical repair. Though an all-autologous repair is favored, the friability of aortic tissue surrounding the pseudoaneurysm made it impossible to achieve a tension-free anastomosis utilizing only native tissue. Thus, we elected to use a homograft patch to reconstruct the aorta at the coarctation segment. While not necessary in this patient, creation of an extra-anatomic bypass via a Dacron graft could be performed via left thoracotomy and can be useful in unstable patients or when the pseudoaneurysm is large.

Unfortunately, our patient rapidly developed recurrent pseudoaneurysm requiring a second operation. Though the median sternotomy approach in the initial surgery offered excellent control for circulatory arrest and coarctation repair, accessing the distal-most portion of the aorta was challenging and recurrence was likely secondary to inadequate source control and debridement. Thus, we would discourage others from using median sternotomy in a similar case.

To optimize access, left posterolateral thoracotomy with left heart bypass was performed for repeat pseudoaneurysm excision. This approach drastically improved visualization and access to the distal aorta, permitting repair without need for circulatory arrest, though left heart bypass was still needed to remove collateral blood flow from the field. In retrospect, the visualization and access the left thoracotomy approach (with/without partial left heart bypass) provided was far superior and would probably be our preferred approach in future similar cases.

Figure 1: CT angiogram demonstrating aortic coarctation A) on admission; B) on hospital day 6 with evidence of pseudoaneurysm development.

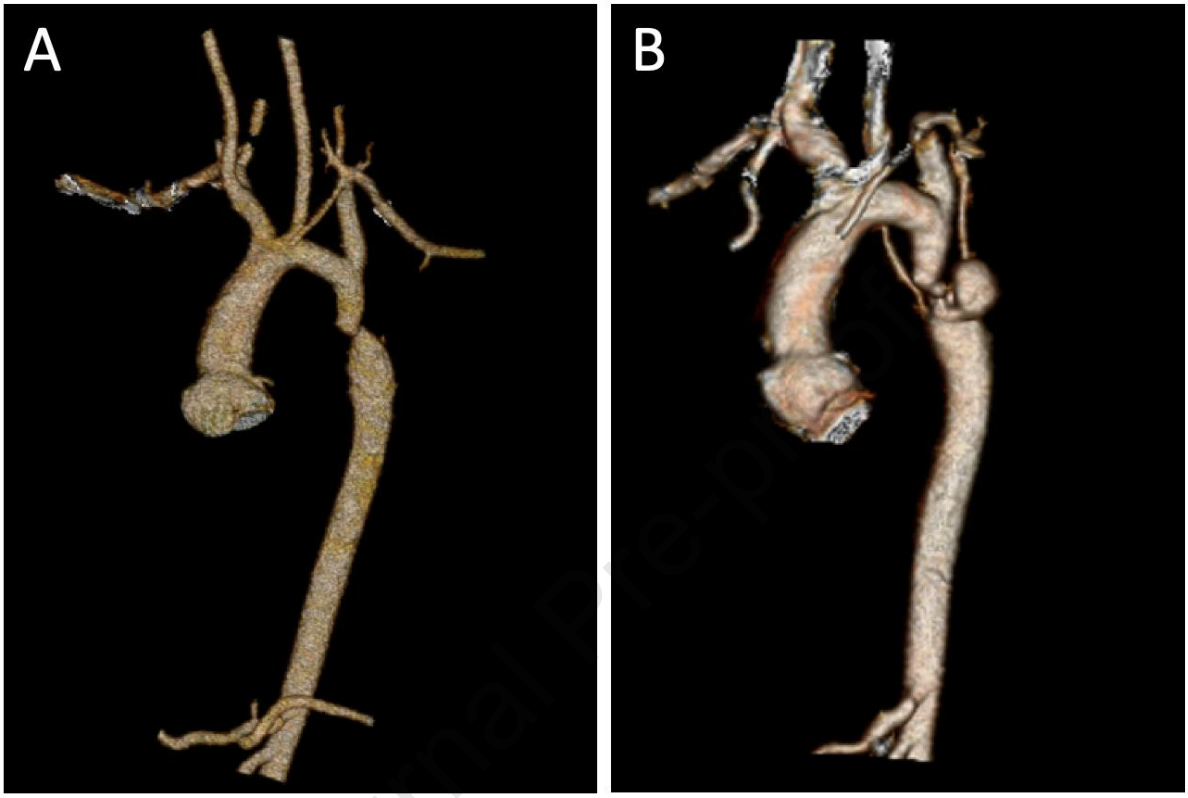


Figure 2: Surgeon's view of aortic coarctation and mycotic pseudoaneurysm via median sternotomy. A) View of mycotic pseudoaneurysm distal to aortic coarctation at initial aortotomy; B) View of coarctation repaired with pulmonary homograft.

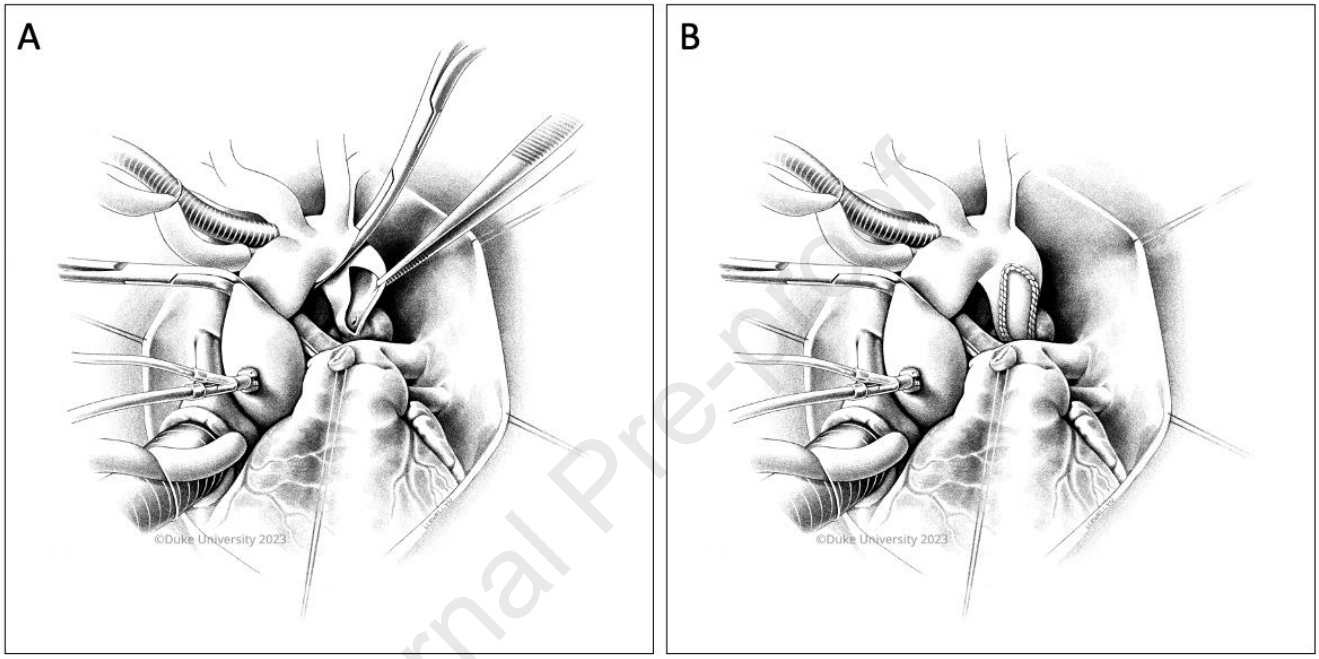


Figure 3: Surgeon's view of recurrent mycotic pseudoaneurysm via left thoracotomy. A) View of recurrent pseudoaneurysm upon entry; B) View of aorta post-repair with aortic homograft.

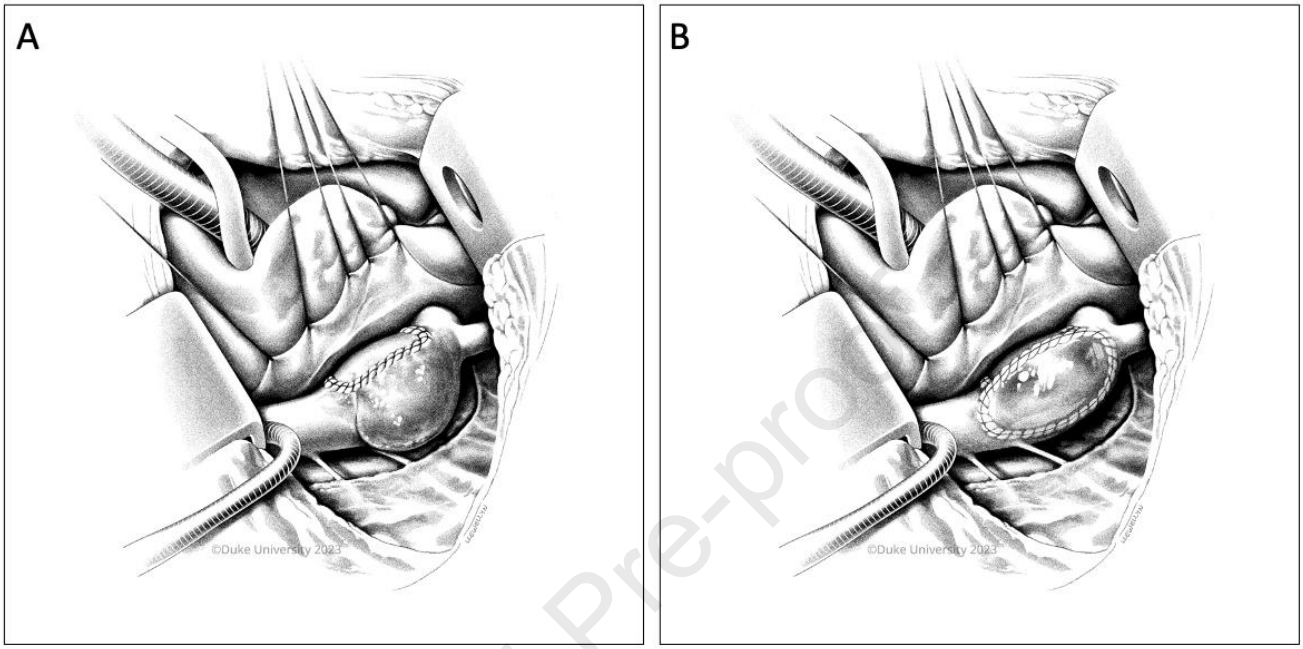
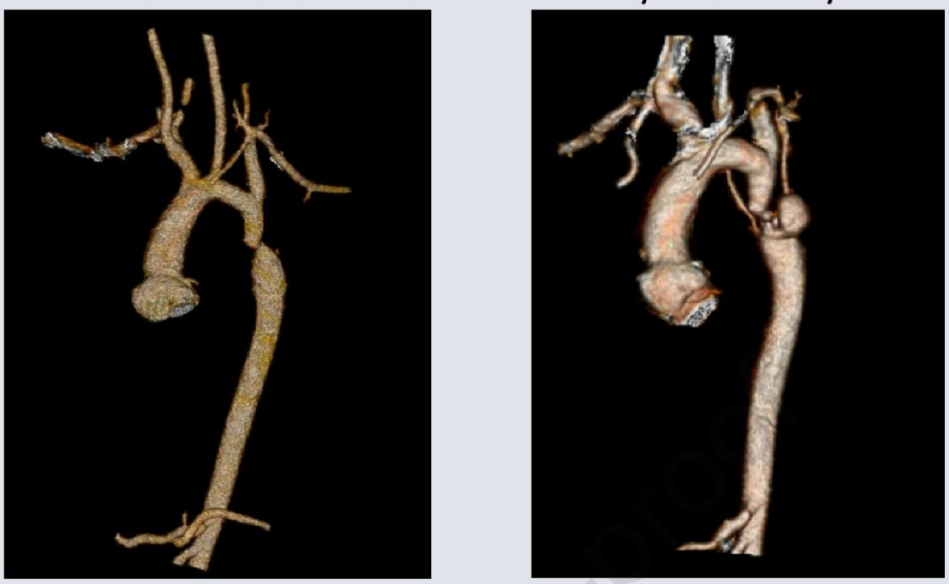


Figure 4: Questions for Consideration

Coarctation of the Aorta with Distal Mycotic Aneurysm



Questions to Consider – Set #1

1. What other imaging would you request?
2. When would you repair the pseudoaneurysm?
3. What is the extent of proximal/distal involvement of the pseudoaneurysm and coarctation?
4. What approach would you use to resect the coarctation and pseudoaneurysm?
5. Would you use bypass or standby? Cooling?
6. Would you patch the defect? If so, with what material?
7. How would you achieve a tension free anastomosis?
8. How long would you continue antibiotic therapy?

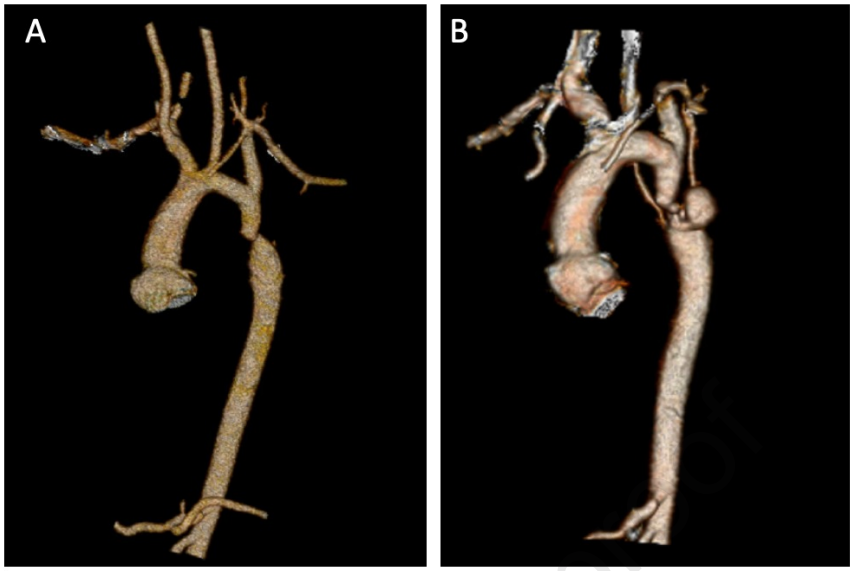
- Uneventful repair via sternotomy with lower body circulatory arrest
- Pseudoaneurysm recurrence during the same hospital admission

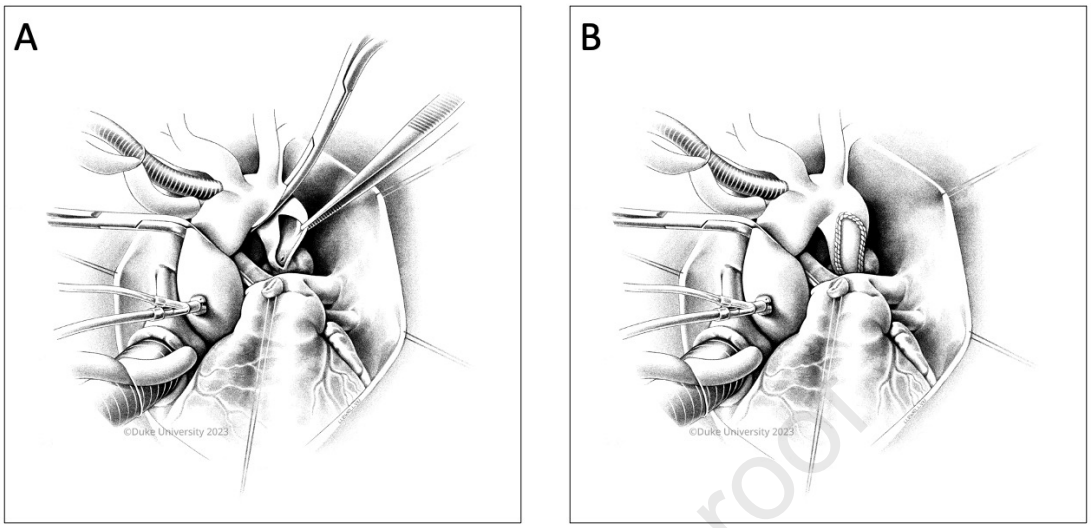
Questions to Consider – Set #2

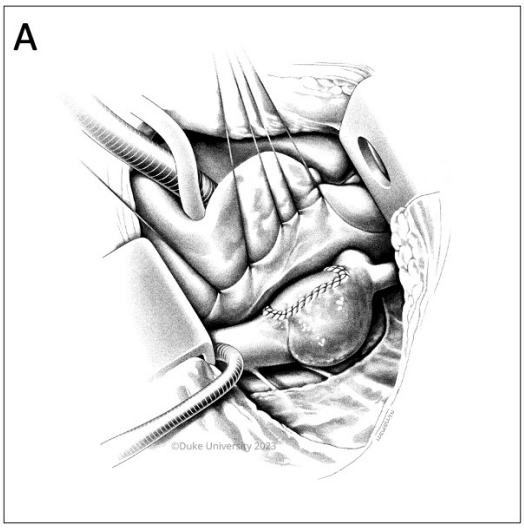
1. Would you discharge this patient home with small recurrent pseudoaneurysm?
2. What follow-up arrangements would you make?
3. What would be indications for reoperation?
4. What approach would you use, and how would you perform the operation?

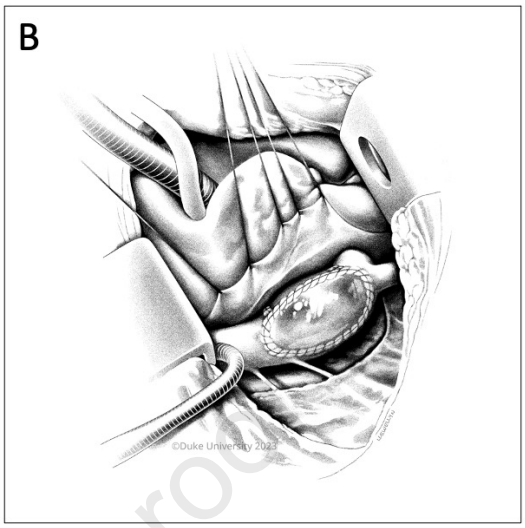
References

1. Ratsimandresy M, Hadeed K, Acar P, Cuttone F. A case report of mycotic pseudoaneurysm in childhood: an unusual complication of coarctation of the aorta. *Eur Heart J Case Rep*. Mar 2019;3(1):yty170. doi:10.1093/ehjcr/yty170
2. Gnanam D, Bartelds B, van Leeuwen WJ, Frohn-Mulder IM, Koopman LP. A case report on endarteritis in a child with coarctation of aorta. *Echocardiography*. Jul 2019;36(7):1427-1430. doi:10.1111/echo.14418
3. Murali S, Wang SS, Grenier MA, Revels JW. "Undiagnosed aortic coarctation with 2 simultaneous acute aortic syndromes: Intramural hematoma and mycotic aneurysm". *Radiol Case Rep*. Oct 2021;16(10):2934-2937. doi:10.1016/j.radcr.2021.07.014
4. Wilson WR, Bower TC, Creager MA, et al. Vascular Graft Infections, Mycotic Aneurysms, and Endovascular Infections: A Scientific Statement From the American Heart Association. *Circulation*. Nov 15 2016;134(20):e412-e460. doi:10.1161/CIR.0000000000000457
5. Grant P, Murala JS, Kolli R, Numa A, Awad J, Dilley AV. Massive hematemesis in a child with undiagnosed aortic coarctation and mycotic aneurysm. *J Thorac Cardiovasc Surg*. Dec 2006;132(6):1482-3. doi:10.1016/j.jtcvs.2006.08.027
6. Tomar M, Parakh R, Bhan A. Mycotic aneurysm: a lesser known complication of coarctation repair in a child, managed by endovascular aneurysm repair. *Images Paediatr Cardiol*. 2017;19(4):1-9.
7. Hafez H, Jamjoom R, Yuones E, Khan Z, Ashour M. Endovascular repair of a mycotic thoracic aortic aneurysm in a patient with aortic coarctation. *J Vasc Surg Cases*. Jun 2015;1(2):154-156. doi:10.1016/j.jvsc.2015.04.008









Coarctation of the Aorta with Distal Mycotic Aneurysm

Questions to Consider – Set #1

1. What other imaging would you request?
2. When would you repair the pseudoaneurysm?
3. What is the extent of proximal/distal involvement of the pseudoaneurysm and coarctation?
4. What approach would you use to resect the coarctation and pseudoaneurysm?
5. Would you use bypass or standby? Cooling?
6. Would you patch the defect? If so, with what material?
7. How would you achieve a tension free anastomosis?
8. How long would you continue antibiotic therapy?

- Uneventful repair via sternotomy with lower body circulatory arrest
- Pseudoaneurysm recurrence during the same hospital admission

Questions to Consider – Set #2

1. Would you discharge this patient home with small recurrent pseudoaneurysm?
2. What follow-up arrangements would you make?
3. What would be indications for reoperation?
4. What approach would you use, and how would you perform the operation?