Journal of Cardiothoracic Surgery

CASE REPORT

Open Access

Pseudoaneurysm of the descending aorta two decades after aortic coarctation repair: a case report

Ruofan Zhou[1†], Yabo Wang[1†] and Qi An[1*]

Abstract

Background Pseudoaneurysm after coarctation of the aorta (CoA) repair is a rare but severe complication. Contributing factors may include infection, hypertension, aortic wall weakness, and turbulent blood flow at the repair site.

Case presentation A 35-year-old male presented with recurrent episodes of epistaxis and dizziness was admitted to the emergency department. He had a history of CoA repair and ventricular septal defect closure 17 years ago. Physical examination revealed elevated blood pressure. Initially, aortic dissection was suspected, but the actual diagnosis was pseudoaneurysm just distal to the left subclavian artery. Surgical intervention involved excision of the pseudoaneurysm and replacement with a new vascular graft via a dual approach of median sternotomy and left thoracotomy. Postoperative recovery was uneventful, and follow-up imaging at one month showed satisfactory aortic morphology.

Conclusions This case underscores the critical role of precise imaging in differentiating pseudoaneurysms from other lesions in post-CoA repair patients. Pseudoaneurysms can present subtly yet carry substantial risks, making regular imaging follow-up essential for early detection and improved outcomes.

Keywords Pseudoaneurysm, Coarctation of the aorta, Aortic dissection, Imaging diagnosis, Complication

Background

Pseudoaneurysm following coarctation of the aorta (CoA) repair represents a rare but severe complication. The reported incidence ranges from 1.3 to 3.0% [1], and it may occur regardless of the surgical technique utilized [2]. The exact cause of pseudoaneurysm formation remains unclear; however, several contributing factors have been suggested, including infection, hypertension, congenital aortic wall weakness, and high-velocity jet streams through the coarcted segment [3, 4]. Here, we present a case of an adult of pseudoaneurysm with a history of CoA repair 17 years prior, who was initially misdiagnosed as aortic dissection (AD).

Case presentation

A 35-year-old male presented to our center with recurrent episodes of epistaxis and dizziness. These symptoms occurred twice—initially during ambulation and later during work activities—and resolved spontaneously each time. Seventeen years prior, the patient had undergone surgical closure of a ventricular septal defect (VSD) via median sternotomy, along with concurrent repair of CoA using a prosthetic bypass through a left thoracotomy in another hospital.

Upon admission, the physical examination indicated elevated blood pressure (BP) measuring approximately 170/82 mmHg, with no significant discrepancy observed between the upper and lower limbs. The nasal endoscopy did not reveal any abnormalities. Computed tomography

†Ruofan Zhou, Yabo Wang have contributed equally to this work.

*Correspondence:
Qi An
anqi@scu.edu.cn
[1] Department of Cardiovascular Surgery, West China Hospital of Sichuan University, 37# Guoxue Xiang, Chengdu 610041, Sichuan, China

angiography (CTA) revealed indications of a concerning AD of the descending aorta (Stanford type B), which aligned with the observation of elevated BP. However, the subsequent image reconstruction corrected the diagnosis as a pseudoaneurysm situated immediately distal to the origin of the left subclavian artery (LSCA), measuring as enlarged as 60 mm (Fig. 1). The pseudoaneurysm presented as a newly formed lumen, rather than the previously implanted prosthetic graft which exhibited significant narrowing. The diameters of the aortic arch and distal descending aorta measured between 15 and 20 mm, with no indication of residual coarctation or AD involved. The electrocardiogram (ECG) showed normal results, ruling out the potential diagnosis of myocardial ischemia (MI). Due to concerns regarding aortic rupture, the decision was made to forgo the exercise test, and the patient was advised to remain on bed rest before surgery.

Following extensive preparation and discussions within a multidisciplinary team (MDT), we opted for median sternotomy combined with left thoracotomy through the fourth intercostal space. Cardiopulmonary bypass (CPB) was set up by cannulating the right femoral artery, the innominate artery, the left common carotid artery, and the right atrium. Intraoperative evaluations confirmed the above findings. The pseudoaneurysm appeared to be associated with anastomotic leakage affecting both the proximal and distal anastomoses of the polyester artificial graft. The graft was anastomosed using continuous Prolene sutures, which were found to be broken for unidentified etiology.

Following isolation of the local adhesion, separate clamps were placed on the right femoral artery, the innominate artery, and the left common carotid artery. Under deep hypothermic circulatory arrest (DHCA) at

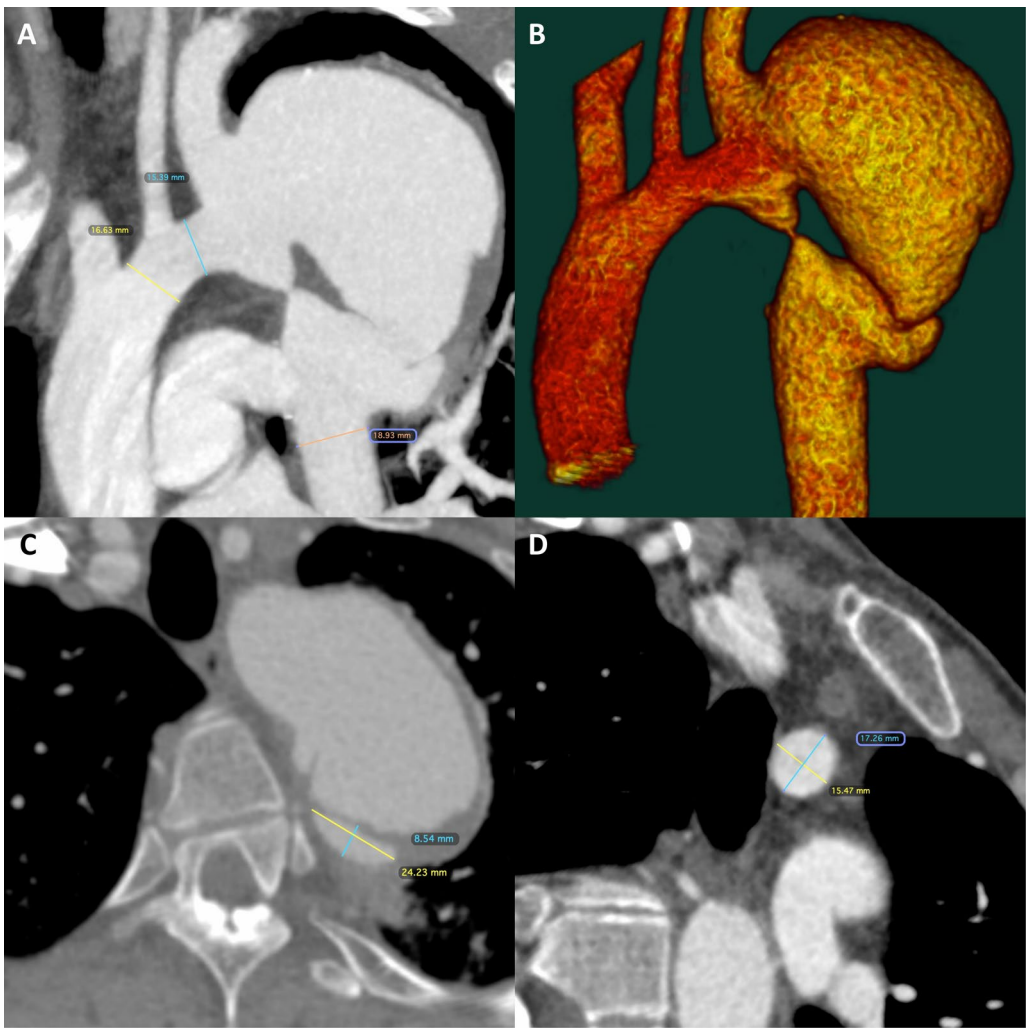

Fig. 1 Preoperative CTA images (**A**, **C**, **D**) and three-dimensional reconstruction (**B**) showing the pseudoaneurysm and aorta

24 °C with antegrade selective cerebral perfusion, the pseudoaneurysm, stenotic artificial bypass graft, and coarcted segment of the aorta were successfully excised.

The minor curvature of the aortic arch and LSCA were slightly longitudinally enlarged. A new 26 mm collagen-coated woven polyester vascular graft was implanted using continuous 4–0 Prolene sutures. A portion of the superior lobe of the left lung was removed because of severe adhesions. The CPB time was 262 min, the cross-clamp time was 180 min, and the selective cerebral perfusion time was 14 min.

The patient was successfully extubated, and postoperative recovery proceeded without complications. A one-month postoperative CTA demonstrated satisfactory morphology of the descending aorta (Fig. 2). Currently, he is under follow-up uneventfully for six months.

Discussion

Previous studies have focused on the predictors of pseudoaneurysm formation following CoA repair. It has been reported that patch aortoplasty using synthetic materials is associated with an elevated risk of pseudoaneurysm or true aneurysm formation [5], with the documented risk up to 21–50% [6]. Additionally, advanced age at the time of CoA repair—specifically beyond a threshold of 13.5 years—has been identified as a potential risk factor [7].

A proposed theory is that the pathology of CoA affects the arterial system as a whole regarding its association with widespread vascular abnormalities [8], such as AD and structural defects in the aorta, especially when accompanied by a bicuspid aortic valve (BAV). Aneurysm may form not only at the site of previous repair, but also in contiguous or remote segments of the aorta, and can also occur in patients without prior surgeries [9]. Notably, intracranial aneurysms are observed in around 10% of patients with CoA, a considerably higher occurrence compared to the general population, as the shared developmental origin of CoA and related vascular anomalies points to a defect in neural crest cells, which contribute to the formation of the aortic arch and other arteries [10]. Therefore, according to this theory, CoA should be regarded as a diffuse arteriopathy rather than a localized abnormality.

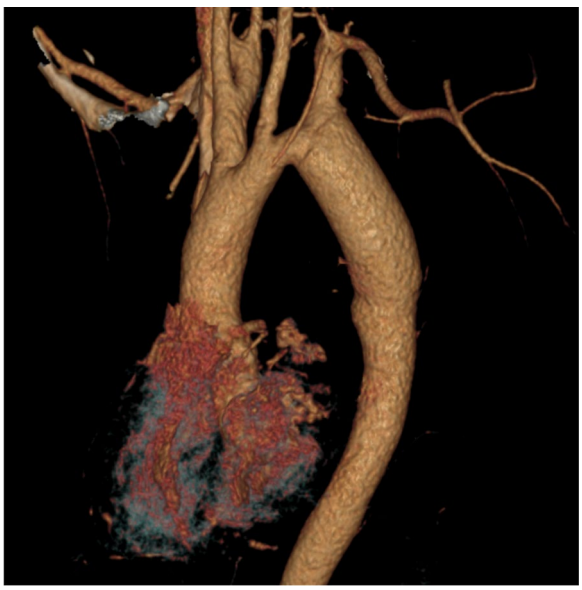

Fig. 2 Three-dimensional reconstruction image displaying the postoperative morphology of the aorta

In the current case, comprehensive preoperative evaluation and therapeutic decision-making are the keys to successfully handling. The patient was initially diagnosed with AD, as the enlarged aorta led by anastomotic leakage and pressed bypass on the cross section mimicking the 'double track sign' including false and true lumens, which is the typical feature of AD. However, the three-dimensional reconstruction demonstrated the actual figure of the great arteries. Following careful analyzing CTA results, the accurate diagnosis as pseudoaneurysm was made.

Surgical strategy is another challenge. Since the detailed documentation of the initial surgical procedure could not be found, it is inferred that the dual-approach strategy was implemented, considering the difficulties of attaining visibility to the CoA in an 18-year-old adult. This aligns with the literature, which indicates that extra-anatomical bypass grafting is generally preferred due to its tension-free nature and relative ease to perform [11, 12]. As for this time, thoracic endovascular aortic repair (TEVAR) was considered optimal initially to minimize trauma, bleeding, as well as complications associated with CPB and DHCA. However, the severe stenosis of the artificial graft made a re-do open operation unavoidable. Considering adhesions and possible collaterals, we decided on a dual approach, rather than isolated sternotomy or left thoracotomy, which provided better intraoperative exposure and saved surgery time.

The onsite was insidious and symptoms of epistaxis and dizziness seemed benign in this case, which is consistent with previous reports that some patients had only mild or even no complaints at all [13]. Consequently, relying solely on symptom-based clinical follow-up is inadequate. Long-term imaging follow-up and surveillance for vascular complications after CoA repair are essential. According to the 2024 European Society of Cardiology (ESC) guidelines [14] and 2022 American Heart Association/American College of Cardiology (AHA/ACC) guidelines [15], all patients post CoA repair should undergo

follow-up with either CTA or magnetic resonance angiography (MRA) every 3–5 years.

Conclusions

Pseudoaneurysm is an uncommon but potentially life-threatening complication following CoA repair. This case underscores the importance of a careful differential diagnosis to distinguish pseudoaneurysm from other conditions like AD, utilizing imaging techniques such as CTA. Given the potential for pseudoaneurysms to present subtly yet carry significant risk, early detection and timely surgical intervention are crucial for achieving favorable outcomes.

Abbreviations

CoA	Coarctation of the aorta
AD	Aortic dissection
VSD	Ventricular septal defect
BP	Blood pressure
CTA	Computed tomography angiography
LSCA	Left subclavian artery
ECG	Electrocardiogram
MI	Myocardial ischemia
MDT	Multidisciplinary team
CPB	Cardiopulmonary bypass
DHCA	Deep hypothermic circulatory arrest
BAV	Bicuspid aortic valve
TEVAR	Thoracic endovascular aortic repair
ESC	European Society of Cardiology
AHA/ACC	American Heart Association/American College of Cardiology
MRA	Magnetic resonance angiography

Acknowledgements

None.

Author contributions

Ruofan Zhou: Conceptualization; Project administration; Writing-original draft. Yabo Wang: Visualization; Writing-original draft. Qi An: Project administration; Supervision; Writing-review & editing.

Funding

None.

Availability of data and materials

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

Ethics approval was waived due to the retrospective nature of this case report. Consent was obtained from the patient's family for participation and publication of anonymized details.

Consent for publication

All authors contributed to and approved the final version of the manuscript. The corresponding author agrees to provide the data underlying this article upon reasonable request.

Competing interests

The authors declare no competing interests.

Received: 16 August 2024 Accepted: 25 December 2024
Published online: 03 January 2025

References

1. Tim Somers et al. Necessity of Life-Long Follow-up after Surgery for Coarctation of the Aorta: A Case Series of Very Late False Aneurysm Formation," ed. Roberto Lorusso et al., European Heart Journal - Case Reports. 2022;6(2): ytac073, https://doi.org/10.1093/ehjcr/ytac073.
2. Perera AH, et al. new-generation stent grafts for endovascular management of thoracic pseudoaneurysms after aortic coarctation repair. J Vasc Surg. 2014;60(2):330–6. https://doi.org/10.1016/j.jvs.2014.02.050.
3. Prifti E, et al. Pseudoaneurysm of the descending aorta complicating an untreated aortic coarctation. Ann Thorac Surg. 2015;99(1):e3-5. https://doi.org/10.1016/j.athoracsur.2014.10.027.
4. Owens CG, et al. Aortic coarctation endarteritis resulting in mycotic pseudoaneurysm and acute mediastinitis. Acta Cardiol. 2010;4:463–5. https://doi.org/10.2143/AC.65.4.2053907.
5. Kotelis D, et al. Endovascular repair of pseudoaneurysms after open surgery for aortic coarctation. Interact Cardiovasc Thorac Surg. 2016;22(1):26–31. https://doi.org/10.1093/icvts/ivv297.
6. Cramer JW, et al. Aortic aneurysms remain a significant source of morbidity and mortality after use of dacron® patch aortoplasty to repair coarctation of the aorta: results from a single center. Pediatr Cardiol. 2013;34(2):296–301. https://doi.org/10.1007/s00246-012-0442-1.
7. Von Kodolitsch Y, et al. Predictors of aneurysmal formation after surgical correction of aortic coarctation. J Am Coll Cardiol. 2002;39(4):617–24. https://doi.org/10.1016/S0735-1097(01)01784-3.
8. Warnes CA. Bicuspid aortic valve and coarctation: two villains part of a diffuse problem. Heart (British Cardiac Society). 2003;89(9):965–6. https://doi.org/10.1136/heart.89.9.965.
9. Preventza O, et al. Coarctation-associated aneurysms: a localized disease or diffuse aortopathy. Ann Thorac Surg. 2013;95(6):1961–7. https://doi.org/10.1016/j.athoracsur.2013.03.062.
10. Rosenquist TH, et al. Impaired elastic matrix development in the great arteries after ablation of the cardiac neural crest. Anat Rec. 1990;226(3):347–59. https://doi.org/10.1002/ar.1092260312.
11. Connery CP, et al. Treatment of aortic coarctation by axillofemoral bypass grafting in the high-risk patient. Ann Thorac Surg. 1991;52(6):1281–4. https://doi.org/10.1016/0003-4975(91)90013-G.
12. Kyohei Hatori et al. Surgical Management of Pseudoaneurysm after Subclavian Artery-to-Descending Aorta Bypass in an Adult Patient with Aortic Coarctation. General Thoracic Cardiovasc Surg Cases 2024;3(1): 26, https://doi.org/10.1186/s44215-024-00154-6.
13. Afifi RO, et al. Redo thoracoabdominal aortic aneurysm repair: a single-center experience over 25 years. Ann Thorac Surg. 2017;103(5):1421–8. https://doi.org/10.1016/j.athoracsur.2016.09.013.
14. Lucia Mazzolai et al. 2024 ESC Guidelines for the Management of Peripheral Arterial and Aortic Diseases. European Heart Journal 2024;45(36): 3538–3700, https://doi.org/10.1093/eurheartj/ehae179.
15. Isselbacher EM, et al. 2022 ACC/AHA Guideline for the diagnosis and management of aortic disease. J Am Coll Cardiol. 2022;80(24):e223-393. https://doi.org/10.1016/j.jacc.2022.08.004.

Publisher's Note