Original Article

Surgical technical experience of adult aortic coarctation concomitant with poststenotic aneurysm or dissection

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Background: Aortic coarctation (COA) in adults combined with poststenotic aneurysm (PA) or poststenotic dissection (PD) is rare and challenging to manage. The existence of multiple factors such as kinking, comorbidities, previous surgical history, and descending aortic lesions increases the difficulty of treatment, and there are currently few clinical reports. The purpose of this study was to present our surgical experience in dealing with such patients.

Methods: A retrospective study was conducted on 20 consecutive patients with COA combined with PA or PD who were treated in our center from December 2015 to April 2019. The basic principles, methods, and short- and mid-term prognosis of surgery are present carefully. This paper introduces the individualized treatment scheme as well as its advantages and disadvantages in detail.

Results: The condition of the included patients was complicated, including 12 cases of PA and 8 of PD. Although different surgical schemes were adopted, procedural success rate was 100%. There were no other surgical complications except 2 cases of anastomotic bleeding and 1 case of spinal cord injury. The results of computed tomography angiography (CTA) demonstrated that 9 cases achieved anatomical correction, 10 cases of PA or PD were eliminated or thrombosed to varying degrees, and only 1 case of PA had no obvious change. Up to the follow-up period, except for 1 patient who had a slight cerebrovascular accident and 1 who had no change in PA underwent cheatham platinum (CP) stent surgery, no other cardiovascular adverse events occurred and all patients recovered well.

Conclusions: The optimal surgical strategy developed collaboratively by cardiac surgeons and endovascular specialists has achieved satisfactory short- and mid-term results for COA patients combined with PA or PD. Further research is still necessary, due to the limited number of cases.

Keywords: Aortic coarctation (COA); poststenotic aneurysm (PA); poststenotic dissection (PD); surgical technical experience

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Introduction

Aortic coarctation (COA) is a common congenital heart disease, occurring in approximately 5–8% of individuals (1). Although the majority of cases are diagnosed and treated during infancy and adolescence, a small number of patients remain undiagnosed until adulthood. These undiagnosed or untreated patients may develop serious complications related to COA, including malignant hypertension, premature coronary artery disease, heart failure, cerebral hemorrhage, sudden death, aortic dissection, or descending aortic aneurysm (2,3). Clinically, adult cases of COA concomitant with poststenotic aneurysm (PA) or poststenotic dissection (PD) are rare, and the treatment is challenging.

In addition to the local anatomy of the COA, other factors should be considered, such as the location, size, and extent of the aneurysm or dissection, the complex collateral circulation network, the presence of proximal coarctation lesions, and rupture of aorta, or the history of previous related cardiovascular surgeries. Previous reports have examined the therapeutic strategy used for these patients, including endovascular (4), surgical (5), and hybrid treatment (6), which have shown positive clinical outcomes. However, most of these reports only focus on simple lesions, lacking comprehensive analysis of the various complicated factors that may influence the outcomes, especially for COA patients combined with PA or PD. In light of this, our article presents the therapeutic experiences and outcomes of 20 patients with different characteristics over the past 5 years. We aim to provide a valuable reference for treating patients with similar conditions appropriately and offer appropriate treatment options. We present this article in accordance with the SUPER reporting checklist (available at https://jtd.amegroups.com/article/view/10.21037/jtd-24-985/rc).

Highlight box

Key findings

- The optimal surgical strategy developed collaboratively by cardiac surgeons and endovascular specialists has achieved satisfactory short- and mid-term results for aortic coarctation (COA) patients combined with poststenotic aneurysm (PA) or poststenotic dissection (PD).

What is known and what is new?

- The existence of multiple factors such as kinking, comorbidities, previous surgical history, and descending aortic lesions increases the difficulty of treatment for COA patients combined with PA or PD, and there are currently few clinical reports.
- This paper details the individualized treatment scheme and its advantages and disadvantages of COA patients combined with PA or PD while stressing the importance of a multidisciplinary team approach.

What is the implication, and what should change now?

- In view of the limited number of patients, a further larger study with long-term follow-up is needed to confirm our results. As new technologies are developed, we will continue to improve the treatment effect of COA patients combined with PA or PD.

Methods

Patients

From December 2015 to April 2019, a total of 131 adult patients (aged ≥18 years) were diagnosed with COA at the Department of cardiac surgery, Beijing Anzhen Hospital, Capital Medical University. All patients underwent computed tomography angiography (CTA) and transthoracic echocardiography (TTE) either at our hospital or other hospitals. PA was defined as a maximum diameter of descending aorta greater than 4.0 cm (7), and PD was defined as the presence of intimal tears and layering. Using these definitions, a total of 20 patients (15.3%) were identified. Baseline information, comorbidities, imaging characteristics, surgical details, and complications were obtained by reviewing electronic medical records. The measurements of the minimum diameter at the site of aortic narrowing and the maximum diameter of the descending aorta were performed by specialized radiologists. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by the Ethics Committee of Beijing Anzhen Hospital (No. 2024131X) and informed consent was taken from all individual participants.

Management strategies and techniques

Compared to the pure COA management strategy, the approach of COA combined with PA or PD are more complex. In addition to considering the comprehensiveness and thoroughness, the procedure's long-term effects should also be considered. The optimal management strategy is determined collaboratively by cardiac surgeons and endovascular specialists. Several principles are primarily considered: (I) the urgency of the condition; when there is an existing arterial rupture or potential risk of rupture, priority is given to a quick and safe approach; (II) the diameter and extent of the aneurysm or dissection, and the location of the intimal tears; (III) the combined congenital heart disease and aortic lesions that require simultaneous treatment; (IV) the complexity of the local and surrounding anatomy of the COA, including the severity of aortic calcification, peripheral collateral circulation (8), and the hypoplastic aortic arch; (V) previous surgical history, such as lateral thoracotomy coarctation correction or median sternotomy cardiovascular surgery; (VI) general clinical factors such as age, physical condition of patients, among others.

Different surgical management strategies are adopted based on the above principles, which are detailed in the following categories.

Aortic replacement, including thoracic aorta replacement (TAR) and thoracoabdominal aortic replacement (TAAR)

This approach is appropriate for patients without congenital heart disease, no history of coarctation correction, and a normal aortic arch proximal to coarctation. Depending on the extent of PA and PD, prosthetic surgical graft replacement is performed through a left thoracotomy or a combined thoracoabdominal incision. If necessary, the left subclavian artery, intercostal artery, and visceral branches need to be reconstructed. Before incising the diseased descending aorta, the hypertrophied and fragile intercostal arteries surrounding the coarctation are carefully dissociated and sutured to prevent rupture and bleeding.

Thoracic endovascular aortic repair (TEVAR)

This intervention is suitable for patients without any abnormalities in the heart, PA diameter less than 45 mm, and PD intimal tear close to the coarctation. An aortic stent graft is placed at the coartcation and PD site using percutaneous vascular access through the common femoral artery, specifically utilizing the cheatham platinum (CP) stent. The pressure changes are assessed at the proximal and distal extermities ofthe stent (aiming for an average post-treatment pressure difference <10 mmHg). Additional balloon dilation and stent remodeling can be performed if the target is not achieved. A stent graft can be used inside the CP stent to ensure complete exclusion of the lesion and increase the coverage area. When the anchoring area is insufficient, the branches of the aortic arch diversion (carotid-subclavian artery diversion, axillary-axillary artery diversion, etc.) can be performed to increase the proximal anchoring area of the stent.

Total arch replacement with elephant trunk surgery with stent grafting (Sun's procedure) (9)

Suitable for young patients with aortic arch dysplasia. Under median sternotomy and deep hypothermic circulatory arrest, a tetrafurcate graft with a specially designed frozen elephant trunk (Cronus®) is used to perform the procedure. To ensure complete sealing of the aneurysm, it is essential to select a frozen elephant trunk with a sufficient diameter. In cases where the stent is not long enough, additional TEVAR intervention can be performed to reconnect the stent and effectively exclude the aneurysm.

Ascending to abdominal aortic bypass (ATAAB)

This surgical approach is suitable for PA combined with congenital heart disease or ascending aorta disease. The procedure involves making a midline thoracoabdominal incision and performing an end-to-side anastomosis of a Dacron prosthetic surgical graft (16 or 18 mm) to the infrarenal abdominal aorta. Cardiopulmonary bypass is established using double-arterial cannulation, with one end placed in the ascending aorta or axillary artery and the other end connected to the prosthetic surgical graft to ensure sufficient systemic perfusion (*Figure 1*). Intravenous cannulation method is depended on whether intracardiac surgery is being performed. The perfusion end of the prosthetic surgical graft is anastomosed to the ascending aorta after completion of the heart or ascending aorta procedure, while the heart is being reanimated. To prevent compression of the gastrointestinal and cardiac areas, the prosthetic surgical grafts are routed behind the gastroduodenum and transverse colon in the abdomen and along the right side of the chest area. The coarctate segment is excluded or ligated, then the distal aneurysm should be closely followed up to observe its thrombosis and whether it has expanded.

Ascending or bisaxillary to femoral artery bypass (ATFAB or BTFAB) and transcatheter closure surgery

This treatment method is for patients with a long segment of PD. The ATFAB involves making an incision in the midline of the chest and both femoral arteries, and using a Y-shaped prosthetic surgical graft. If no thoracotomy is required, two 8-mm prosthetic grafts can be used to perform BTFAB. To avoid any damage to organs caused by the tunnelled device entering the abdominal cavity, the abdominal segment is placed under the skin. After 3 days, transcatheter closure intervention is performed using a ventricular septal defect (VSD) occluder. Anastomosis of both femoral arteries should be anastomosed on a non-dissected location.

Patient follow-up

Clinical follow-up of patients were required to undergo

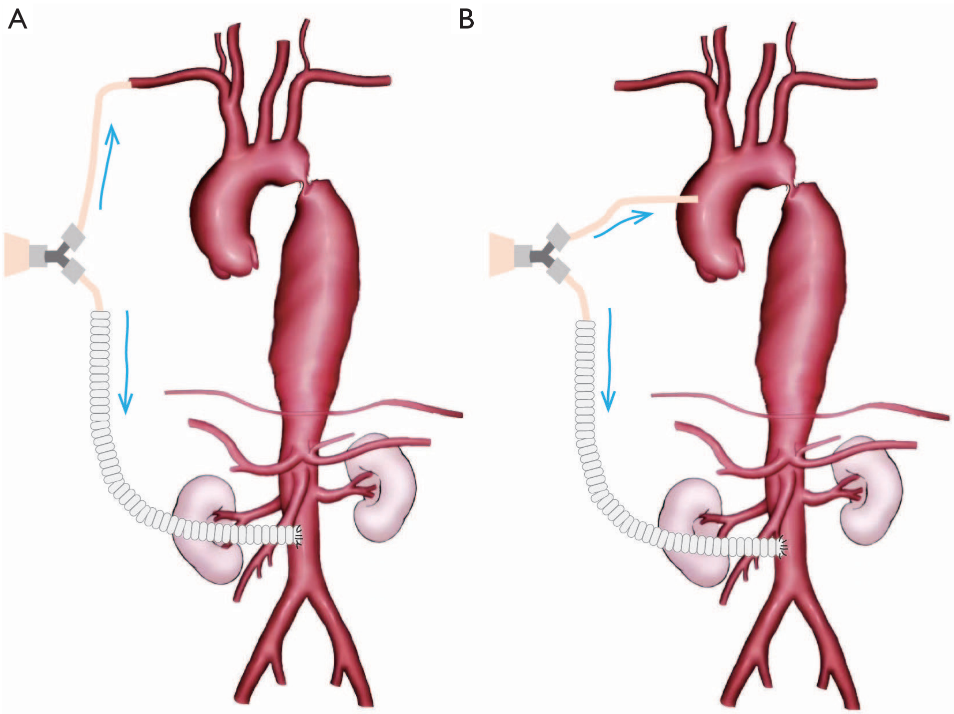

Figure 1 Diagram of double-arterial cannulation, anastomosis of a prosthetic surgical graft to the infrarenal abdominal aorta. (A) The end placed in the ascending aorta for intracardiac disease; (B) another end placed in axillary artery for proximal aorta disease. The blue arrows indicate the direction of perfusion blood flow during cardiopulmonary bypass.

aortic CTA to evaluate aortic changes 1 year after surgery. Comprehensive follow-up of all patients was conducted in October 2023, encompassing patient survival, re-intervention, and complications. The follow-up results were obtained through electronic communication with patients.

Results

Among the enrolled 20 patients, 12 had PA, and 8 had PD. The average age at surgery was 43 years (19–64 years), with 17 patients (85.0%) being male. A total of 16 patients (80%) had hypertension as a complication. Among these hypertension patients, 1 (5.0%) had COPD, 2 (10.0%) had coronary heart disease but did not meet the surgical criteria, and 1 (5.0%) experienced ischemic stroke without sequelae. All patients experiencing PD reported severe chest and back pain upon onset. There were 2 PA patients (aneurysm diameter >50 mm) who presented with mild chest pain, whereas the rest had no chest pain. The surgical procedures were all successfully carried out with meticulous decision-making by the surgical team. The average postoperative follow-up time was 74 months (range, 56–96 months), with no deaths.

Complete anatomical correction was achieved in 9 patients, including 5 TAR or TAAR, 3 TEVAR, and 1 Sun's procedure (*Table 1*). One patient who underwent thoracic aortic replacement required an emergency thoracotomy to address a massive anastomotic hemorrhage. Fortunately, this patient did not experience any postoperative complications related to the cardiovascular system, nervous system, or infection and recovered smoothly. The remaining patients also recovered well without any surgery-related complications. Postoperative aortic CTA scans for all patients indicated successful treatment of the coarctation segment, aneurysms, and dissections. The prosthetic surgical grafts were in good condition, with no pseudoaneurysm formation at the anastomosis (*Figure 2*). The covered stent effectively sealed the dissection tears without any endoleakage, and the aortic wall remodeling went well (*Figure 3*). A small amount of contrast agent was observed around the stent end in the patient who underwent Sun's surgery, prompting strict follow-up (*Figure 4*). As of the latest follow-up, there have been no re-operations or adverse events related to the aorta or heart.

Among another 4 patients with PD (*Table 2*), 3 (No.

Table 1 Procedural details of patients undergoing anatomical correction

Case	Lesion type	CD (mm)	DAD (mm)	Comorbidities	Operation method
1	PD	5	58	AVR	TAAR (22×10×8×8×10 mm prosthetic graft)
2	PD	8	42	None	TAR (22 mm prosthetic graft)
3	PA	9	58	None	TAR + LSAR (22×10×8×8×10 mm prosthetic graft)
4	PA	4	50	BAV; normal function	TAR (24 mm prosthetic graft)
5	PA	7	55	None	TAR (32 mm prosthetic graft)
6	PA	11	41	None	TEVAR (3.9 cm CP stent)
7	PD	6	54	Aortic arch anatomical variation	TEVAR (4.5 cm CP stent + 30×30×200 mm covered stent)
8	PD	8	56	Aortic arch hypoplasia	Axillary-axillary artery diversion (8 mm prosthetic graft) + TEVAR (30×30×200 mm covered stent + 32×32×150 mm covered stent)
9	PA	8.4	53	Aortic arch hypoplasia	Sun's procedure (30 mm prosthetic graft + 8 mm prosthetic graft + 14 mm prosthetic graft + 30×100 mm frozen elephant trunk)

PD, poststenotic dissection; PA, poststenotic aneurysm; CD, coarctation diameter; DAD, descending aorta diameter; AVR, aortic valve replacement; BAV, bicuspid aortic valve; TAAR, thoracoabdominal aortic replacement; TAR, thoracic aorta replacement; LSAR, left subclavian artery reconstruction; TEVAR, thoracic endovascular aortic repair; CP, cheatham platinum.

Figure 2 Pre- and post-operative imaging of patient No. 1. (A) Preoperative CTA demonstrating that the range of dissection extends to the renal artery; (B) postoperative CTA demonstrating complete elimination of constricted segment and dissection. CTA, computed tomography angiography.

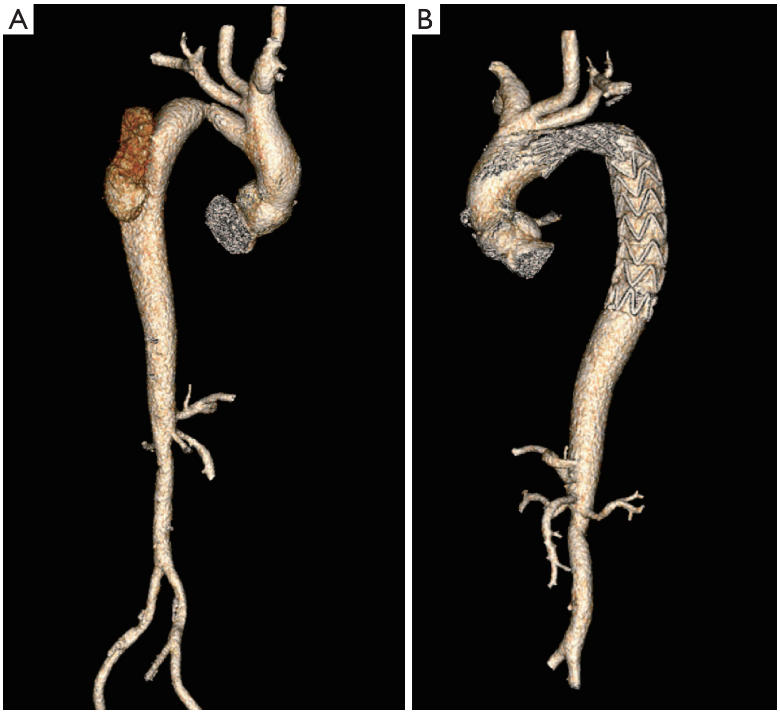

Figure 3 Pre- and post-operative imaging of patient No. 7. (A) Preoperative CTA demonstrating bovine aortic arch combined with COA and the dissection is limited, the intimal tear is located in the proximal of the descending aorta; (B) postoperative CTA demonstrating complete aortic remodeling. CTA, computed tomography angiography; COA, aortic coarctation.

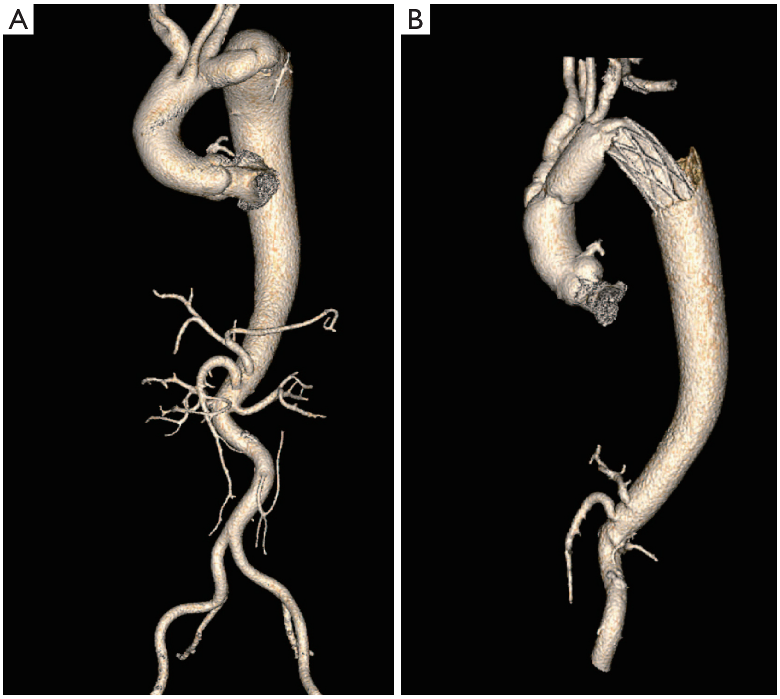

Figure 4 Pre- and post-operative imaging of patient No. 9. (A) Preoperative CTA demonstrating twisted coarctation; (B) postoperative CTA demonstrating normal arch anatomy and contrast agent was observed around the stent end. CTA, computed tomography angiography.

Table 2 Procedural details of patients concomitant with PD

Case	Lesion type	CD (mm)	DAD (mm)	Comorbidities	Operation method
10	PD	7	40	None	ATFAB (16×8×8 mm Y prosthetic graft) + transcatheter closure
11	PD	6.9	40	None	ATFAB (18×9×9 mm Y prosthetic graft) + transcatheter closure
12	PD	4	45	BAV; aortic valve stenosis	AVR (22 mm mechanical valve) + ATFAB (16×8×8 mm Y prosthetic graft) + transcatheter closure
13	PD	9	41	Aortic root aneurysm	Bentall + (25 mm mechanical valved tube) + ATAAB (16×30 mm, 14×15 mm prosthetic graft)

PD, poststenotic dissection; CD, coarctation diameter; DAD, descending aorta diameter; BAV, bicuspid aortic valve; ATFAB, ascending to femoral artery bypass; AVR, aortic valve replacement; ATAAB, ascending to abdominal aortic bypass.

Figure 5 Pre- and post-operative imaging of patient No. 11. (A) Preoperative CTA demonstrating that the dissection is extensive, reaching to iliac artery; (B) post-repair CTA demonstrating the proximal descending aorta completely thrombosed. CTA, computed tomography angiography.

10–12) had dissections involving the descending aorta, abdominal aorta, and iliac arteries, which were successfully treated with transcatheter closure intervention following ATFAB. The remaining 1 patient with dissection that extended to the celiac trunk underwent ATAAB, avoiding transcatheter closure, because the coarctation segment was completely occluded upon post-ATAAB angiography. All patients recovered and were discharged, and no complications such as spinal cord injuries, visceral vascular ischemia, or anastomotic bleeding occurred. The last CTA examination showed that the prosthetic graft was unobstructed and coursed naturally; the proximal descending aorta or false lumen was completely thrombosed, and the residual dissected segment was not dilated (*Figure 5*). Throughout the follow-up period, only patient No. 13 experienced a minor cerebral hemorrhage due to inadequate warfarin adjustment, whereas the remaining three patients were in excellent health.

Table 3 Procedural details of patients concomitant with PA

Case	Lesion type	CD (mm)	DAD (mm)	Comorbidities	Operation methods
14	PA	9	40	Ascending aortic dissection	Ascending aorta replacement (28 mm prosthetic graft) + ATAAB (16×30 mm prosthetic graft)
15	PA	4	41	Aortic valve stenosis; mitral valve regurgitation	Bentall (23 mm mechanical valved tube) + ATAAB (16×30 mm prosthetic)
16	PA	4	50	Mitral valve regurgitation	MVR (28 mm mechanical valve) + ATAAB (14×30 mm prosthetic graft)
17	PA	11	48	None	ATAAB (16×30 mm prosthetic graft)
18	PA	3	40	Calcifications of coarctation	ATAAB (16×30 mm prosthetic graft)
19	PA	8	45	Aortic valve regurgitation	AVR (23 mm mechanical valve) + ATAAB (18×30 mm prosthetic graft)
20	PA	9	52	None	ATAAB (18×30 mm prosthetic graft) + constrictive segment ligation

PA, poststenotic aneurysm; CD, coarctation diameter; DAD, descending aorta diameter; ATAAB, ascending to abdominal aortic bypass; MVR, mitral valve replacement; AVR, aortic valve replacement.

The remaining 7 patients who underwent ATAAB all had PA, and 4 of them underwent cardiac or aortic surgery simultaneously (*Table 3*). Patient No. 20 presented with a small diameter artery of the aortic arch proximal to the coarctation, posing challenges in matching the proximal and distal ends of the prosthetic surgical graft for arterial replacement surgery, thus necessitating bypass surgery. During the procedure, the coarctation segment was easily exposed, prompting an attempted ligation of the coarctation segment. Unfortunately, post-operatively, the patient experienced numbness in both lower limbs and weakened muscle strength, attributed to spinal cord injury resulting from intercostal artery ischemia due to rapid thrombosis of the descending aorta. After 24 hours of cerebrospinal fluid drainage and enhanced anticoagulation, the patient's condition returned to normal. There were no symptoms of spinal cord injury in the other patients. Except for patient No. 14 who underwent thoracotomy for hemostasis due to bleeding from the proximal anastomosis of the prosthetic surgical graft, no other surgery-related complications were observed. Clinical symptoms of internal organ or intestinal compression by prosthetic surgical graft were absent. Postoperative aortic CTA revealed complete exclusion of the aneurysm in patient No. 20, with partial thrombosis in the distal part of the descending aorta (*Figure 6*). Similar changes were noted in other patients except for patient No. 15, where the aneurysm nearly disappeared, normal blood flow was observed on the medial side of the descending aorta lumen, and thrombosis formed on the lateral side (*Figure 7*). Throughout the follow-up period, there were no adverse events such as cardiac, nervous system, or gastrointestinal accidents, and all patients experienced successful recovery. There were no instances of distal vascular embolism due to intra-aortic thrombus dislodgement. Patient No. 15 underwent CP stenting in another hospital 24 months after surgery because the aneurysm remained unchanged.

Discussion

The underlying mechanisms leading to aortic aneurysm or dissection caused by COA may be that the blood flow velocity at the coarctation site is accelerated, and the high-speed blood flow impacts the distal arterial wall continuously for a long time. The arterial wall structure changes occur as a consequence (10) and can even progress towards dilatation or rupture. A study showed that about 21% of patients with untreated coarctation died of aneurysm rupture (11). A case of ruptured aortic pseudoaneurysm associated with COA during pregnancy admitted to our center survived after timely and successful treatment (12). Therefore, once COA with PA or PD is found, we should intervene as early as possible to avoid adverse consequences.

For isolated COA with PA or PD without congenital heart disease, prosthetic surgical graft replacement and endovascular stent implantation (13,14) can achieve anatomical correction of such patients, which aims to restore physiologic hemodynamic characteristics of the

Figure 6 Pre- and post-operative imaging of patient No. 20. (A) Preoperative CTA, a small diameter artery at the front arch of the coarctation and PA; (B) post-repair CTA, aneurysmal disappearance and partial thrombosis in the distal part of the descending aorta. CTA, computed tomography angiography; PA, poststenotic aneurysm.

human body and is the ideal therapeutic approach. The traditional management relies on open surgery with vascular replacement, providing an effective COA treatment but is highly invasive and carries the risk of hemorrhage. Interventional procedures with angioplasty and stent grafts can solve this problem well. For patients with insufficient anchorage area at the proximal end of stent, the branches of the aortic arch can be diverted to increase the anchorage area. Pan *et al.* (15) also introduced the same treatment strategy and achieved good short-term results, but stent leakage and restenosis also have to be considered (16).

For patients with PA associated to COA and who need concomitant correction of congenital heart disease, there is a dilemma in choosing the treatment sequence. Treating the cardiac disease priority may put the aneurysm at risk of rupture, while treating the aneurysm first may lead to perioperative heart failure. Staged treatment, on the other hand, can increase trauma and economic burden. Bypass surgery is an alternate option (17,18), and whether to ligate or putting-aside the coarctation is decided according to the difficulty of treatment. From this study, both methods can achieve the purpose of thrombosis and prevention of aneurysm rupture, but the thrombosis effect of ligation is more accurate, yet it is necessary to be alert to the occurrence of spinal cord ischemia in the early stage. Distal vascular embolism caused by thrombus shedding in the aneurysm did not appear in this group. In addition, further follow-up is needed to determine whether the thrombotic aneurysm will continue to expand in the long term. Generally speaking, several patients with aneurysms in this group did not expand in the short- and mid-term.

Compared with aneurysms, dissection treatment is more complicated; besides considering the extension of the lesion, we also need to consider the intimal tear location and the influence of true and false lumen on important branches. Therefore, we developed a new hybrid treatment approach consisting of two separate steps: first, we performed bypass surgery with a prosthetic surgical graft (ATAAB). Then, 3 days later, we performed endovascular vaso-occlusion using a VSD occluder device to seal the coarctation site.

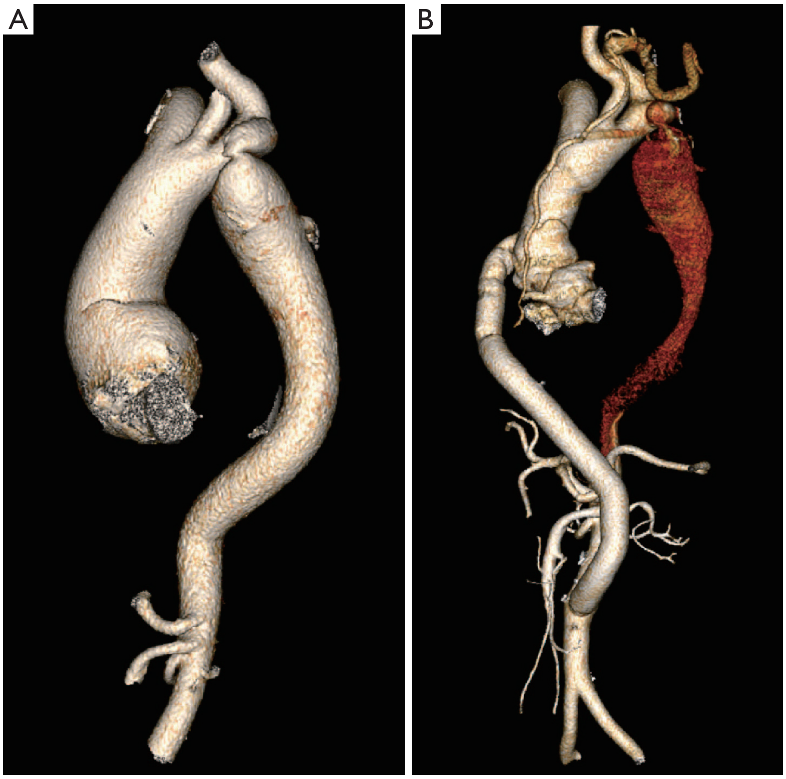

Figure 7 Pre- and post-operative imaging of patient No. 14. (A) Preoperative CTA showing ascending aorta dissection and PA; (B) post-repair CTA showing normal blood flow in the medial side of the descending aorta and thrombosis formed in the lateral side. CTA, computed tomography angiography; PA, poststenotic aneurysm.

This method also avoided the need for balloon pre-expansion of the arterial wall at the coarctation site, diverting the blood flow path, and achieved the purpose of thrombosing the false lumen (19). There was no difficulty in making the guidewire cross the constricted segment, and in this group of patients, the approach was proven to be safe. However, this method is similar to the traditional type B aortic dissection interventional procedure, and cannot seal distal re-entry tears, enabling progressive false lumen dilatation and warranting further follow-up (20).

The present study is limited by its retrospective design and the relatively small patient cohort, despite being relatively large for COA with PA and PD. Due to the low incidence of this rare disease, it is challenging for a single center to enroll a sufficient number of patients to generate statistically significant results. Additionally, the majority of patients are not native to Beijing, and the long-term CTA results are difficult to obtain, which is also a major limitation. In order to provide reliable technical reference for these rare patients, we will continue to collect accumulating case data, obtain more long-term efficacy evaluation, and develop new surgical techniques to improve the therapeutic effect.

Conclusions

Overall, building on traditional surgery, our center combines the advantages of both surgical and endovascular therapy. This approach incorporates both well-established approach and innovation, enabling individualized decision-making for this group of patients through teamwork, and has achieved satisfactory short-term and medium-term results. Further research is still necessary, due to the limited number of cases.

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Footnote

Reporting Checklist: The authors have completed the SUPER reporting checklist. Available at https://jtd.amegroups.com/article/view/10.21037/jtd-24-985/rc

Data Sharing Statement: Available at https://jtd.amegroups.com/article/view/10.21037/jtd-24-985/dss

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://jtd.amegroups.com/article/view/10.21037/jtd-24-985/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by the Ethics Committee of Beijing Anzhen Hospital (No. 2024131X) and informed consent was taken from all individual participants.

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