

## Hybrid Repair of Late-Onset Pseudoaneurysm After Previous Surgical Coarctation Repair

AYŞE ZEHRA KARAKOÇ,<sup>1</sup> MUSTAFA AKBULUT,<sup>2</sup> ADNAN AK,<sup>2</sup> MEHMET ALTUĞ TUNCER<sup>3</sup>

<sup>1</sup>Department of Cardiovascular Surgery, Kırıkkale High Specialization Hospital, Kırıkkale, Türkiye

<sup>2</sup>Department of Cardiovascular Surgery, University of Health Sciences, Kosuyolu High Specialization Training and Research Hospital, İstanbul, Türkiye

<sup>3</sup>Department of Cardiovascular Surgery, Okan University Hospital, İstanbul, Türkiye

### Abstract

Coarctation of the aorta is a congenital malformation that has long been considered completely correctable with appropriate surgery in childhood. Late thoracic pseudoaneurysms develop in 5–12% of patients undergoing repair for coarctation of the aorta, the incidence varying with the method of repair when the first time that coarctation has been diagnosed. Two cases have been reported in this article. The first patient who is a 35-year-old male had the saccular pseudoaneurysm which sized 6.5×7.5 cm placed in the aortic arch and included the left common carotid artery which occurred as a late-onset complication after open end-to-end repair of the coarctation. A hybrid repair such as left carotid-subclavian bypass in the operation room and after then thoracic endovascular aortic repair intervention in the angiography room was compromised by the surgical team. The second patient who is 30-year-old male had an aortic aneurysm which sized 6.2×7.2 cm placed in the descending aorta that was originated from next to the orifice of the left subclavian artery. He had a history of aortic coarctation and underwent open surgical repair 14 years prior. The surgical procedure initiated with an exploration of the right axillary artery then a median sternotomy was performed. Each of the supra-aortic branches is prepared. The aortic valve was bicuspid and normofunctional. The aneurysm was resected up to the aortic root, followed by a proximal anastomosis using a 28 mm Dacron graft. Subsequently, the cross clamp was placed on the brachiocephalic artery. Then, the frozen elephant trunk procedure was performed. The left subclavian artery anastomosed above the separated graft using 10 no dacron graft. The patient weaned from cardiopulmonary bypass without any problems, stayed in the ICU for 2 days, and discharged 2 weeks after the operation. Hybrid treatment combines endovascular intervention and extra-anatomic bypass as carotid-subclavian bypass performed in this case successfully. However, these procedures; both of them can be very challenging and they carry their own potential pitfalls; pre-operative planning with a whole surgical team makes this safer and easier.

**Keywords:** Aneurysm repair; aortic coarctation; pseudoaneurysm.

## Koarktasyonun Cerrahi Tamiri Sonrası Gelişen Geç Başlangıçlı Psödoanevrizmalarda Hibrit Tedavi

### Özet

Aort koarktasyonu onarımı geçiren hastaların %5 ila %12'sinde, koarktasyonun ilk kez teşhis edildiği yöntemle bağlı olarak değişen insidanslarla, geç torakal psödoanevrizmalar gelişebilir. Bu makalede iki hasta sunulmuştur. İlki, 35 yaşında bir erkekti ve koarktasyon onarımı sonrası geç dönemde ortaya çıkan 6,5×7,5 cm boyutlarında sakküler psödoanevrizmaya sahipti. Bu psödoanevrizma, aort arkında bulunan ve sol ortak karotis arteri de içermekteydi. Cerrahi ekibi tarafından işlem odasında sol karotid-subklavyen bypass gibi bir hibrit onarım ve ardından anjiyografi odasında TEVAR gerçekleştirildi. İkinci hasta, 30 yaşında bir erkekti ve sol subklavyen arter bitiminden orijinlenen 6,2×7,2 cm boyutlarında bir desendan aort anevrizmasına sahipti. Bu hasta, aort koarktasyonu teşhisi almış ve 14 yıl önce açık cerrahi onarım geçirmişti. Cerrahi prosedür, önce sağ aksiller arterin incelenmesi ile başladı, ardından median sternotomi uygulandı. Supraaortik dalların her biri hazırlandı. Aort kapak biküspit ve normofonksiyoneldi. Anevrizma, aort köküne kadar rezekte edildi, ardından 28 numara Dacron greft kullanılarak proksimal anastomoz gerçekleştirildi. Daha sonra kros klemp brakiosefalik artere yerleştirildi ve donmuş fil hortumu prosedürü

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### Address for Correspondence:

Ayşe Zehra Karakoç

Department of Cardiovascular Surgery,  
Kırıkkale High Specialization Hospital,  
Kırıkkale, Türkiye

**E-mail:** aysezehra@gmail.com

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uygulandı. Sol subklavyen arter, ayrılmış greftin üzerine 10 numara Dacron greft kullanılarak anastomoze edildi. Hasta, kardiyopulmoner baypasstan sorunsuz bir şekilde çıkarıldı ve 2 gün yoğun bakımda kaldıktan sonra 2 hafta sonra taburcu edildi. Her iki vakada da kombine hibrit tedaviler başarıyla tamamlanmıştır. Her ne kadar prosedürel zorluklar olsa da preoperatif cerrahi ekip tarafından detaylı değerlendirme ile bu iki yaklaşımın da geç başlangıçlı psödoanevrizma tamerinde başarılı ve güvenli olabileceği gösterilmiştir.

**Anahtar sözcükler:** Anevrizma onarımı; aort koarktasyonu; psödoanevrizma.

## Introduction

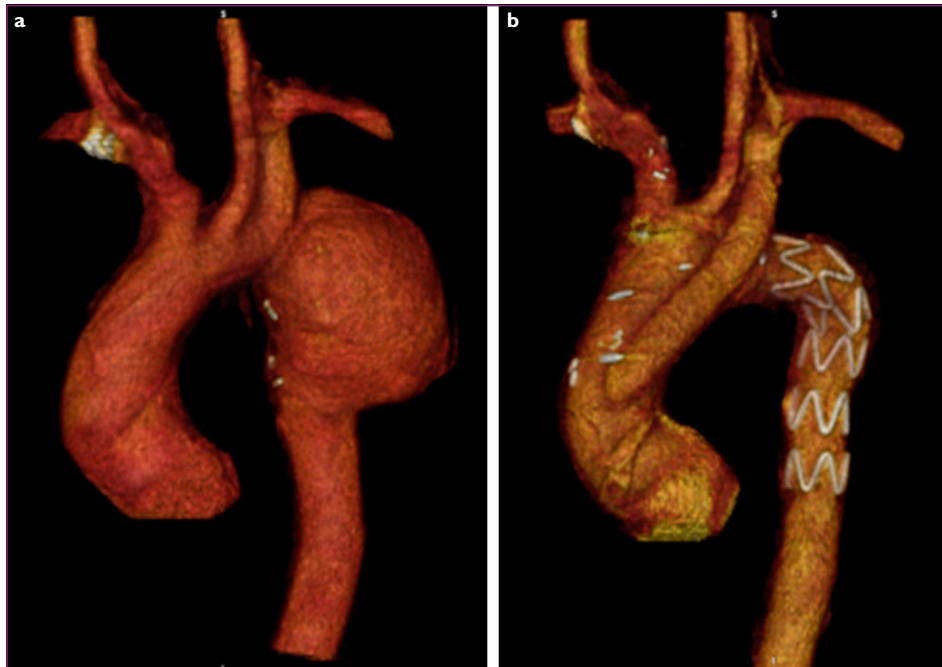
Coarctation of the aorta is a congenital malformation that has long been considered completely correctable with appropriate surgery in childhood. However, with the aging of these patients, many late-onset complications have been reported such as systemic hypertension, aneurysms, recurrent coarctation, and aortic valve dysfunction.<sup>[1]</sup> The late-onset pseudoaneurysms after surgical repair were observed in all various methods; such as subclavian flap angioplasty (17%), patch angioplasty (14%), tube graft repair (6%), or end-to-end anastomosis (3%).<sup>[2]</sup> Besides high pre-operative peak systolic pressure gradients and bicuspid aortic valve are predictive risk factors for the development of post-surgical pseudoaneurysm.<sup>[3]</sup> Whatever the surgical technique used in the first repair, redo open surgical repair for pseudo-aneurysms has high mortality rates.<sup>[4]</sup> However, hybrid repair has been reported as safe and effective.<sup>[3]</sup> We report two different hybrid repairs of coarctation-associated aneurysm: Left carotid-subclavian bypass with thoracic endovascular aortic repair (TEVAR) and supra-coronary aortic replacement and aorta left subclavian bypass with open stent grafting (OSG).

## Case Report

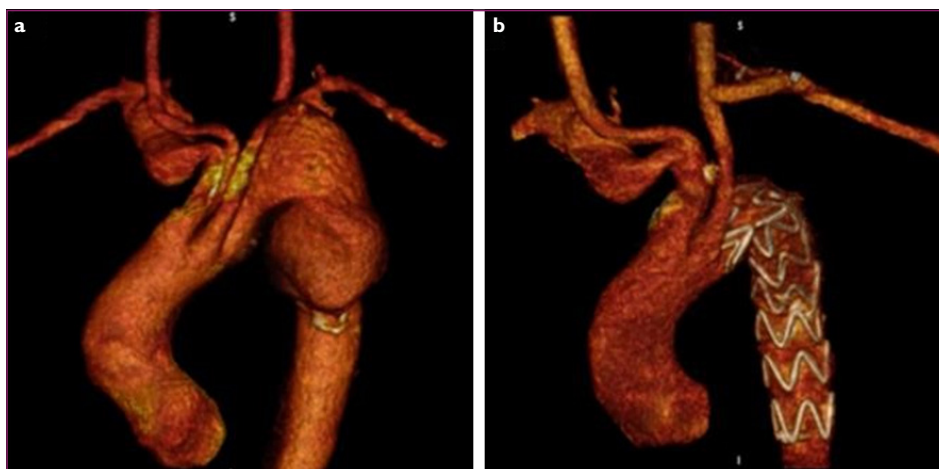
### Case I

A 35-year-old man was admitted to the cardiovascular surgery clinic with dyspnea and hoarseness which started 5–6 months

ago and increased rapidly in a few days. He had a history of aortic coarctation repair by patch angioplasty 18 years ago. A computed tomography angiogram (CT-A) signified the saccular pseudoaneurysm which sized 6.5×7.5 cm placed in the aortic arch and includes the left subclavian artery. The aortic arch was 21 mm and the descending aorta distal to the aneurysm was measured as 23 mm. A control echocardiogram revealed a normal tricuspid aortic valve and preserved left ventricular ejection fraction of 65%. Since the suitable 2 cm proximal landing zone for endovascular repair could be provided with a left carotid-subclavian bypass, the surgical team decided to perform a hybrid arch repair. Extra anatomic bypass was performed as bilateral carotid-subclavian bypasses using 7 no Dacron grafts through supraclavicular incisions under general anesthesia and the proximal left subclavian artery was surgically ligated proximal to the left vertebral artery ostium. Then, endovascular repair from the right common femoral artery as retrograde fashion to zone-2 was performed with a 26×26×200 mm TEVAR stent-graft (Bolton Medical [Sunrise, USA], Relay® Plus). Completion angiography confirmed the correct position of the endograft without evidence of endoleak. Before discharge, control CT-A was performed after the session without wasting time but no significant endoleaks were noted. He was discharged without any post-operative complications or clinical problems. He had no problems in both imaging and the 1-year follow-up (Fig. 1).



**Figure 1.** Case I. Volume rendering and 3D reconstruction (a) before and (b) 1 year after left carotid-subclavian bypass with thoracic endovascular aortic repair.



**Figure 2.** Case 2. Volume rendering and 3D reconstruction show a pseudoaneurysm (a) before and (b) 2 years after supra-coronary aorta replacement, aorto-left subclavian bypass with open stent grafting.

## Case 2

A 30-year-old man was admitted to our hospital with back pain for 3 months. He had a history of aortic coarctation repair by aorto-aortic bypass with a 20-mm vascular graft 14 years ago. A saccular aortic aneurysm which sized 6.2×7.2 cm placed in the descending aorta that originated from next to the orifice of the left subclavian artery was detected in CT-A. Furthermore, he had 46 mm in a proximal site of the ascending aorta. The echocardiogram revealed that the left ventricular ejection fraction was 65%, the bicuspid aortic valve was normofunctional, and ascending aorta was 45 mm. Considering the anatomical features, age, and surgical risks, we decided to perform OSG with the repair of the ascending aorta. Central catheter, bilateral upper extremity arterial monitorization, near-infrared spectroscopy, and cerebrospinal fluid pressure catheter were placed. While the right subclavian artery was used for arterial cannulation, venous drainage was provided using the right atrium. A vent cannula was placed from the upper-right pulmonary vein. Myocardial protection was ensured with blood cardioplegia. The aortic valve was bicuspid and normofunctional without any morphological pathology. Proximal aorta repair was performed with a handmade one-branched graft (28 no+10 mm Dacron graft) in the cooling phase. When the nasopharyngeal temperature was 26°C, the aortic clamp was removed, and bilateral antegrade selective cerebral perfusion (flow rate=10–15 mL/kg/min) was started. Then, OSG was performed using tapered relay NBS plus stent graft (200-mm length with proximal and distal diameters of 20 mm and 24 mm, respectively, Bolton Medical; Sunrise, USA) to the Zone-2 through pre-placed guidewire. The stent graft was fixed to the aorta wall through interrupted U sutures at the proximal of left carotid artery. Afterward, distal anastomosis was completed with hemi-arch replacement, and warming was initiated. Antegrade selective cerebral perfusion time was 77 min. The aorta-left subclavian artery bypass was performed using a previously implanted 10 no Dacron graft. Without any post-operative complications and clinical problems, the patient was discharged. At 1 and 2-year follow-up visits, CT-A scans showed complete aneurysm exclusion and the patient was fully asymptomatic (Fig. 2).

## Discussion

Late-term aortic pseudoaneurysm formation after surgical repair of aortic coarctation has been reported for all repair methods including patch aortoplasty, end-to-end anastomosis, and prosthetic graft replacement.<sup>[2]</sup> Pseudoaneurysm structure has a high risk of spontaneous rupture rate of 31% and rupture-related mortality is high consequently.<sup>[4]</sup> Redo open surgical repair has a high risk of bleeding, pulmonary complications, and renal complications with high mortality.<sup>[2]</sup> Hybrid treatment can be the solution for difficult cases including concomitant cardiac diseases and complications of re-thoracotomy. In both cases, there are technical considerations for our choice of endovascular repair in different hybrid procedures: The length of the segment to be covered, the length of the suitable proximal landing zone, and the mismatch in the proximal and distal landing zones. The first case is a standard hybrid arch repair and a suitable proximal landing zone was created by performing an extra-anatomic bypass such as left carotid-subclavian bypass. However, the Bolton Relay Plus stent graft was specifically chosen to overcome the tortuosity of the aortic arch since it is more flexible after exiting the outer primary sheath. In the second case, a median sternotomy was required for two reasons: (1) The management of the 4.6 cm ascending aorta of a young patient with bicuspid aortic valve and coarctation and (2) the need to create an adequate landing zone for standard TEVAR in young patients, which can only be achieved with a left carotid-subclavian bypass at a maximum of 1.1 mm, so debranching is required. Furthermore, the surgical team decided to apply supra-coronary replacement, aorto left subclavian bypass, and OSG for pseudoaneurysm treatment since OSG has the advantage of providing fixation of the proximal side of the stent-graft even with a 1 cm suturing space. In addition, there was a mismatch in the aortic diameter before and after the pseudoaneurysm. We performed proximalization of the stent-graft landing zone using an aorta-left subclavian bypass, along with antegrade placement of the tapered Relay Plus TEVAR stent graft. This approach effectively addressed the

mismatch problem. The longest diameter of the standard frozen elephant trunk hybrid grafts available in the market is 150 mm long. To properly cover the lesion, we employed a 200 mm long stent graft in the OSG procedure.

## Conclusion

Our report shows that hybrid repair is an encouraging solution for pseudo-aneurysms following open surgery for aortic coarctation. Moreover, no matter how complex aortic pathology is, hybrid repair can be used safely with appropriate device preferences and detailed pre-operative planning.

## Disclosures

**Informed Consent:** Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

**Authorship Contributions:** Concept – A.Z.K., M.A.; Design – A.Z.K., M.A., A.A., M.A.T.; Supervision – A.Z.K., M.A., A.A., M.A.T.; Funding – A.Z.K., M.A., A.A., M.A.T.; Materials – A.Z.K., M.A., A.A., M.A.T.; Data collection and/or processing – A.Z.K., M.A., A.A., M.A.T.; Data analysis and/or interpretation – A.Z.K., M.A., A.A., M.A.T.; Literature search – A.Z.K., M.A., A.A., M.A.T.; Writing – A.Z.K., M.A., A.A., M.A.T.; Critical review – A.Z.K., M.A., A.A., M.A.T.

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