Complex Hybrid Repair of an Aberrant Right Subclavian Artery With Rapidly Developing Kommerell's Diverticulum

Kavaliunaite, Egle; Kjeldsen, Bo Juel; Midtgaard, Annette; Akgul, Cengiz

Published in:
EJVES Vascular Forum

DOI:
10.1016/j.ejvsvf.2023.11.003

Publication date:
2024

Document version:
Final published version

EJVES Vascular Forum (2024) 61, 12—15

CASE REPORT

Complex Hybrid Repair of an Aberrant Right Subclavian Artery With Rapidly Developing Kommerell's Diverticulum

Egle Kavaliunaite [a,*], **Bo Juel Kjeldsen** [a], **Annette Midtgaard** [b], **Cengiz Akgul** [a]

[a] Department of Cardiac, Thoracic, and Vascular Surgery, Odense University Hospital, Odense, Denmark
[b] Department of Interventional Radiology, Odense University Hospital, Odense, Denmark

Introduction: An aberrant right subclavian artery (ARSA) is an aortic anomaly that, in some cases, can be complicated with Kommerell's diverticulum (KD) at the origin of the ARSA. Progression and rupture of KD are associated with high mortality. Timely intervention is therefore required; however, there are no clinical guidelines for the most suitable intervention.
Report: A 50 year old, previously healthy, male patient developed dysphagia. He was diagnosed with an aberrant right subclavian artery and KD. The KD increased in size from 4 — 7 cm within 2 months. He underwent single stage hybrid aortic repair involving an aortic valve replacement, total aortic arch debranchment, two thoracic endovascular aortic repair stents, and subclavian plugs. He developed a stroke during the post-operative period; however, all neurological symptoms had disappeared at 6 months and computed tomography showed no endoleaks and all supraortic vessels were open.
Discussion: Literature on KD is limited; therefore, there is no consensus on KD treatment. Increasing awareness of rapidly developing KD will add to current knowledge of the disease. One stage cardiac and non-cardiac surgery was successfully performed with no long term complications.

Article history: Received 12 June 2023, Revised 16 October 2023, Accepted 8 November 2023,
Keywords: Aberrant right subclavian artery, Aortic arch repair, Arteria lusoria, Kommerell diverticulum

INTRODUCTION

An aberrant right subclavian artery (ARSA) with a left-sided aortic arch is one of the most common aortic abnormalities, with a prevalence of 0.4% in the general population.[1] Between 20 — 60% of cases involving the ARSA are linked to the presence of an aneurysmal formation at the origin of the ARSA, which is referred to as Kommerell's diverticulum (KD). Most cases are asymptomatic and about 5% may present with symptoms, the two most common being dysphagia due to compression of the oesophagus and tracheal compression.[2] Kommerell's diverticulum might manifest by rupture or dissection, which may occur in 15—53% of cases.[3]

Due to the rarity and anatomical variety of these cases, there are no well established guidelines or consensus for treatment. The surgical treatment options include traditional surgical open repair, thoracic endovascular aortic repair (TEVAR), and hybrid operation. An increasing number of publications have reported various successful hybrid techniques for the treatment of KD over the past decade,[4] providing an alternative to open repair.

This report describes a complex hybrid case involving total aortic arch debranching, a biological aortic valve replacement, thoracic stent grafting (TEVAR), and subclavian plugs in a patient with ARSA and KD.

CASE

A 50 year old white male with no prior history of cardiovascular diseases complained of severe dysphagia and was referred to the vascular surgery department after a routine chest radiograph showed enlargement of the upper mediastinum, suggesting the presence of an aortic arch anomaly. Computed tomography angiography revealed a left side aortic arch with an aberrant right subclavian artery with KD, which had increasingly grown from 4 to 7 cm in 2 months. Upon further investigation, the patient also had an aneurysmatic sinus of 5.7 cm and severe aortic valve insufficiency with a reduced left ventricular ejection fraction (LVEF) of 35%.

The multidisciplinary committee determined a hybrid procedure with full aortic debranching and biological aortic valve operation to be most appropriate in this case. Although the standard treatment option for this condition is usually a frozen elephant trunk operation, this option was rejected due to the patient's poor LVEF and concomitant need for

* Corresponding author. Odense University Hospital, J. B. Winsløws Vej 4, 5000, Odense, Denmark.
E-mail address: egle.kavaliunaite@rsyd.dk (Egle Kavaliunaite).

https://doi.org/10.1016/j.ejvsvf.2023.11.003

aortic valve replacement. It was decided to shorten the time on the heart—lung machine because it required a cooling down temperature of 20 oC for a patient with LVEF of 35% and severe aortic valve insufficiency (Fig. 1).

Surgery

The operation started with a cutdown in the right groin while simultaneously dissecting bilateral infraclavicular subclavian arteries underneath the clavicle. A Dacron prosthesis was sewn into the right subclavian by end to side anastomosis using 5—0 Prolene. Afterward, a 10 mm prosthesis was sewn to the right femoral artery. Perfusion was obtained via a heart—lung machine.

A median sternotomy was performed, the pericardium was opened, and cannulation was performed when the activated clotting time was >400 seconds. Firstly, the heart—lung machine was connected to the right femoral artery and the right subclavian artery, then a two-staged venous cannula was inserted into the right atrium. Antegrade and retrograde cardioplegia were inserted, and dissecting around the aorta was performed. A non-biodegradable polytetrafluoroethylene (PTFE) felt was placed around the aorta and an aortic clamp was placed on it. Cardioplegia was initiated and supplemented every 20 minutes. The aorta was opened, the aortic root dissected, and "buttons" were formed for both coronary arteries. It was noticed that the left ostia divided into circumflex and left anterior descending arteries at the aortic wall.

The aortic valve was excised, and the valve size was measured. A bioprosthetic Perimount Magna Ease 29 aortic valve (Edwards Lifesciences, Irvine, CA, USA) was placed inside at the end of a 32 mm vascular prosthesis (Gelweave size 32 Vascutek Terumo Corporation, Glasgow, UK) and fixed to the aortic annulus using Ethibond 2—0 Prolene with a PTFE felt piece placed on the ventricular side. Then, the coronary "buttons" were re-inserted into the graft with running 5—0 Prolene sutures. Finally, the Dacron aortic prosthesis was used to make an end to end anastomosis with the distal part of the ascending aorta close to the aortic arch. The heart was then de-aired, the aortic clamp was removed, and reperfusion and mechanical ventilation were started.

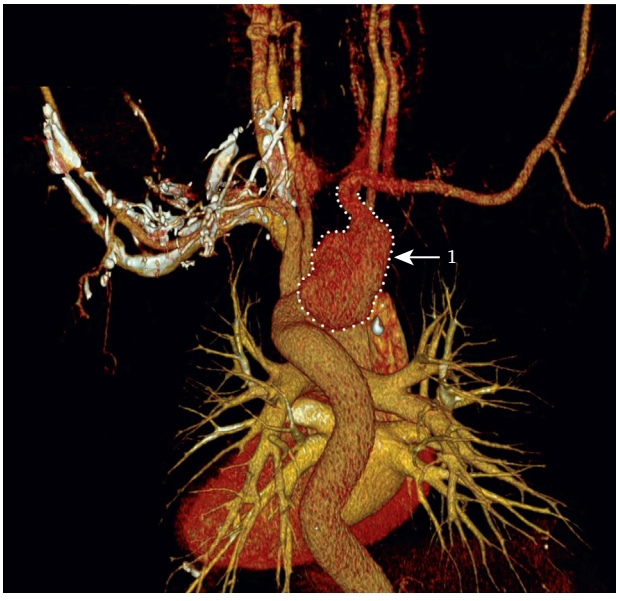

Figure 1. Pre-operative three dimensional computed tomography reconstruction showing Kommerell's diverticulum (white arrow) in a dorsal view of the aortic arch.

Debranching

Total aortic debranching began with the proximal end to side anastomosis of the trifurcated graft being sewn end to side to the ascending aorta using a 4—0 Prolene suture. After this, the focus was on the distal anastomosis: the left infraclavicular space was dissected and the first branch of the trifurcated graft was sewed end to side to the left subclavian artery using a 5—0 Prolene suture. The second branch of the graft was sewed to the left common carotid artery end to side using 5—0 Prolene and proximal to this anastomosis the artery was ligated with Ethibond. The last third branch was sewn end to side to the right common carotid artery, and this was proximally ligated like the second branch. A PTFE graft was then tunnelled to the right infraclavicular space, and this graft was sewn into the right subclavian artery.

Once the debranching was performed, endovascular repair followed. Access was gained through the femoral artery. Two stents (Medtronic, Valiant, 97 x 34 mm, Minneapolis, MN, USA) were placed in the aortic arch with landing zone 0.

Lastly, by puncturing the right subclavian anastomosis for access, two plugs (Amplatzer, 18 x 18 mm, Abbott, Chicago, IL, USA) were inserted into the right subclavian artery proximal to the vertebral artery, to avoid backflow to the aneurysm, and adequate flow to the vertebral artery was ensured by the retrograde flow. Similarly, two plugs were inserted proximal to the vertebral artery in the left subclavian artery. Thus, the bilateral vertebral arteries were preserved. Control angiography confirmed complete exclusion of the aneurysm and KD with no endoleaks and confirmed all supraortic bypasses to be open (Figs. 2 and 3).

Post-operative course

The patient was then transferred to the intensive care unit, where he suffered a stroke on post-operative day 3 that resulted in left hemiparesis and vision loss. The patient made a quick recovery and was discharged to rehabilitation with minimal vision loss and reduced strength of the left arm. After 6 months, he had regained full vision and strength in his left arm and was able to receive his driver's license back. Control computed tomography scans after 1 and 2 years show no endoleaks and all supraortic vessels were open. The patient had no post-operative complaints.

DISCUSSION

There is no consensus on ARSA or KD treatment criteria because the prevalence of this condition is low. Upon the

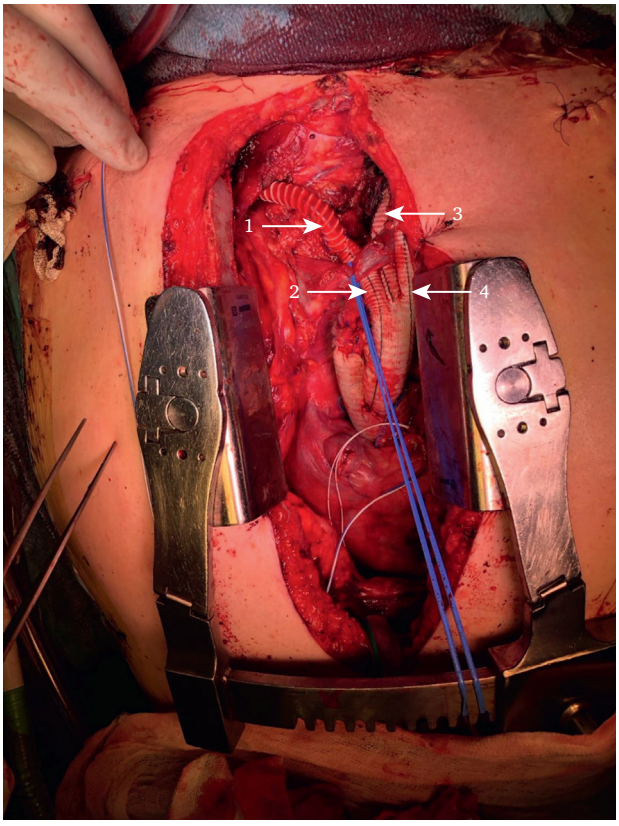

Figure 2. Trifurcated aortic arch prosthesis with right subclavian artery bypass. 1. Right subclavian artery; 2. Right common carotid artery; 3. Left common carotid artery; 4. Left subclavian artery.

accidental finding of KD, some centres prefer to delay intervention, arguing that slow progression of KD does not pose a significant risk of rupture. A retrospective study investigated 75 KD cases and reported annual growth of KD 1.45 ± 0.39 mm/year, thus supporting the previously mentioned practice.[5]

Although the size at which these aneurysms could rupture cannot be predicted, some argue that treatment should only be offered to symptomatic patients, while others offer repair when KD diameter reaches 30 or 50 mm in distance to the opposite aortic wall.[6] This means that patients are offered control scanning upon reaching this size.

This case provides new insights into KD, as KD developed from 4 to 7 cm in 2 months, suggesting a potentially dangerous growth rate. Rapidly developing aneurysms always require the possibility of infection to be explored; this suspicion was refuted as the patient did not show any clinical signs of infection (no fever, normal inflammatory markers) and no signs of infection on the computed tomography scan. Rupture is one of the most fatal complications arising from KD and is strongly associated with high mortality. Cinà et al. published a series and review in which 32 patients (53%) presented with rupture and dissection, thus encouraging early intervention.[7] The literature on KD treatment is still limited and no consensus on treatment criteria exists, so it is vital to individually evaluate the risk of rupture, even in cases of small KD.

Open repair through thoracotomy has been widely used as the conventional treatment approach for ARSA with KD, although it has a relatively high mortality rate of 8%.[8] In cases with a right sided aortic arch connected to a left descending aorta, the trachea overrides the aortic arch, and the estimated surgical risk is extremely high. The recent advancement in minimally invasive endovascular TEVAR has become an attractive option for selected patients, as it helps to avoid the morbidity of a thoracotomy or sternotomy incision and the need for aortic cross-clamping or deep hypothermic circulatory arrest. Although TEVAR is associated with lower mortality, it also poses other issues such as complications due to the main branches of the arteries being isolated. A recent systematic review assessed the current treatment strategies for the management of ARSA and KD to compare treatment outcomes and complication

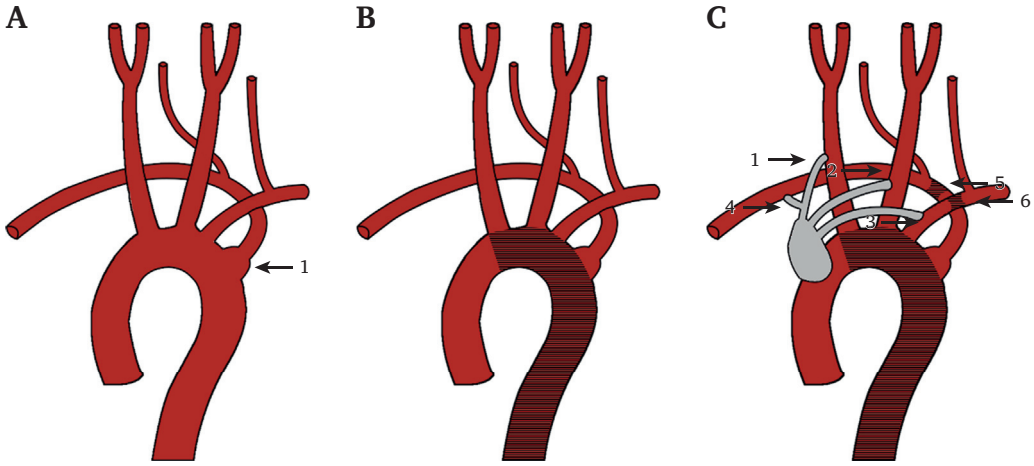

Figure 3. Schematic visualisation of the procedure. (A) Pre-operative anatomy (1. Kommerell's diverticulum); (B) Thoracic endovascular aortic repair landing zone 0; (C) Trifurcated graft (1. Bypass to the right carotid artery; 2. Bypass to the left carotid artery; 3. Bypass to the left subclavian artery; 4. Bypass to the right subclavian artery; 5. Vascular plugs in the right subclavian artery; 6. Vascular plugs in the left subclavian artery).

rates. When the authors of the study compared postoperative stroke complications between open surgery, endovascular repair, and the hybrid approach, insignificant between group differences were reported.[9] Moreover, other studies have reported higher incidences of stroke in TEVAR patients in ascending aneurysms when compared with descending aneurysms.[10] A full endovascular approach is only appropriate in cases without an atrial septal aneurysm, otherwise the origin of the aneurysm can only be successfully ligated using thoracotomy. This was not a technically viable option for the current patient, as he was not a candidate for an operation with prolonged time on a heart—lung machine and cooling down to 20 oC. Thus, the treatment approach involved open total debranching, TEVAR with embolisation, and aortic valve operation. The vertebral arteries were preserved by placing two plugs in the right and left subclavian prevertebral, and circulation was ensured by retrograde flow.

REFERENCES

1 van Son JAM, Konstantinov IE. Burckhard F. Kommerell and Kommerell's diverticulum. *Tex Heart Inst J* 2002;**29**:109—12.

2 Austin EH, Wolfe WG. Aneurysm of aberrant subclavian artery with a review of the literature. *J Vasc Surg* 1985;**2**:571—7.

3 Singh S, Chen JF, Assi R, Nassiri N, Vallabhajosyula P. Intraoperative retrograde TEVAR to control endoleak after emergent total arch replacement and frozen elephant trunk repair for ruptured Kommerell's diverticulum. *J Card Surg* 2020;**35**:3578—80.

4 De Rango P, Cao P, Ferrer C, Simonte G, Coscarella C, Cieri E, et al. Aortic arch debranching and thoracic endovascular repair. *J Vasc Surg* 2014;**59**:107—14.

5 Erben Y, Brownstein AJ, Velasquez CA, Li Y, Rizzo JA, Mojibian H, et al. Natural history and management of Kommerell's diverticulum in a single tertiary referral center. *J Vasc Surg* 2020;**71**:2004—11.

6 Tanaka A, Milner R, Ota T. Kommerell's diverticulum in the current era: a comprehensive review. *Gen Thorac Cardiovasc Surg* 2015;**63**:245—59.

7 Cinà CS, Althani H, Pasenau J, Abouzahr L. Kommerell's diverticulum and right-sided aortic arch: a cohort study and review of the literature. *J Vasc Surg* 2004;**39**:131—9.

8 Siregar S, Groenwold RHH, de Mol BAJM, Speekenbrink RGH, Versteegh MIM, Bravo Brandon, Bruinsma GJ, et al. Evaluation of cardiac surgery mortality rates: 30-day mortality or longer follow-up? *Eur J Cardio-thorac Surg Off J Eur Assoc Cardio-thorac Surg* 2013;**44**:875—83.

9 Loschi D, Santoro A, Rinaldi E, Bilman V, Chiesa R, Melissano G. A systematic review of open, hybrid, and endovascular repair of aberrant subclavian artery and Kommerell's diverticulum treatment. *J Vasc Surg* 2023;**77**:642—9.

10 Melissano G, Tshomba Y, Bertoglio L, Rinaldi E, Chiesa R. Analysis of stroke after TEVAR involving the aortic arch. *Eur J Vasc Endovasc Surg Off J Eur Soc Vasc Surg* 2012;**43**:269—75.