



Case Report

Hybrid Repair for Acute Aorto-Pulmonary Fistula Following Coarctation Repair

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Abstract

Pseudoaneurysm is an infrequent complication of aortic coarctation repair. It occurs in 5-9% of patients. Further complications such as pseudoaneurysm rupture or aorto-pulmonary fistula (APF) carry a high mortality rate and represent a therapeutical and surgical challenge. A 42-year-old man, with a history of 2 prior aortic coarctation repairs at age 5 and 16, presented to our emergency department with persistent hemoptysis for one week. Angio-scan showed a bulky thoracic aortic pseudoaneurysm measuring 79 x 69 mm localized at the aortic isthmus. The pseudoaneurysm was associated with peri-aneurysmal infiltration of the lung parenchyma, highly suggestive of an aorto-bronchopulmonary fistula. The patient underwent urgent hybrid repair, with thoracic stentgraft in zone I (E-vita, Jotec[®]) and retropharyngeal right Common Carotid Artery (CCA) to left subclavian and left carotid arteries artery Dacron bypass graft. Post-operative course was uneventful. After total recovery, the patient was discharged at post-operative day 5. There are no current guidelines for the management of this rare complication. Multidisciplinary discussion is mandatory to identify the best options. In this case, patient was successfully treated with hybrid repair without pulmonary resection.

Introduction

Pseudoaneurysm is an infrequent but life-threatening complication of aortic coarctation repair. It occurs in 5-9% of patients and, left untreated, carries poor medium-term prognosis. Further complications such as pseudoaneurysm rupture or Aorto-Pulmonary Fistula (APF) carry a high mortality rate and are a therapeutic as well as a surgical challenge, not least because they require immediate attention, accurate diagnosis and decisive action. The standard management approach of such complications is open surgery. Yet, this can become particularly delicate because these complications tend to occur in patients who already underwent prior open surgery in this location, and because most such patients are still young, having undergone the initial surgery in infancy or in their teenage years. This might partly explain that the intra-operative mortality during open surgical management is reported to be non-negligible (around 15%) [1]. Furthermore, the infrequent occurrence of such complications makes them rarities,

best managed by a multidisciplinary team, and to this day devoid of generally agreed-upon guidelines. In this case report, we wish to present our choice of a hybrid management technique, which proved surgically adequate and clinically successful.

Case Report

A 42-year-old man was transferred to our hospital following several episodes of hemoptysis that progressively increased over a week and were accompanied by cough and dyspnea (NYHA class 2). He reported neither chest pain nor palpitations and had no known exposure to tuberculosis. The patient had undergone two aortic coarctation repairs, 26 and 37 years earlier. Regular follow-up every other year was managed by his cardiologist. The patient was not under medication, had no history of drug abuse, never smoked and had no family history of systemic diseases. At initial presentation, the patient was hemodynamically stable and his cardiovascular evaluation showed no sign of respiratory distress.

The laboratory analyses yielded results for hemoglobin level at 160g/l (normal range: 133-177 g/l) and white blood cell count at 10.7 G/l (normal range: 4.0-10.0 G/l). Cardiac enzymes were all within normal range. The chest X-ray showed a widening of the mediastinum and multidetector contrast-enhanced Computed Tomography (CT) demonstrated a bulky (79 x 69 mm) thoracic aortic pseudoaneurysm at the aortic isthmus starting 7 mm distally to the left subclavian artery origin and ending 6 cm lower (Figure 1a-c). Examination of the lung window revealed peri-aneurysmal infiltration in the upper left lung parenchyma with multiple pulmonary ground glass opacities in the apex (Figure 1d). Because of the radiological signs of alveolar hemorrhage, the presence of the bulky aneurysm and the clinical presentation, we suspected an aorto-bronchopulmonary fistula, a rare complication of aortic pseudoaneurysms. Because the patients had a history of two prior thoracic surgeries, and because of the proximal localization of the

pseudoaneurysm, a multidisciplinary decision was made to exclude the aneurysm using a thoracic endograft that would ultimately cover the left CCA and subclavian arteries. The surgical procedure was performed using a transverse incision over the anterior border of the right sternomastoid, with the patient positioned in dorsal decubitus to expose the right CCA. A second left supraclavicular incision was made to expose the left CCA. The dissection of the left subclavian artery was carried out all the way to the origin of the vertebral and internal mammary arteries. To establish the bypass, a 7mm Dacron tube was connected end-to-side to the right CCA and end-to-end to the Left Subclavian Artery (LSA) proximally to the vertebral and to the internal mammary arteries. The prosthetic bypass was tunneled in the retropharyngeal and retroesophageal space. Figure 2 shows the reimplantation of the left CCA on the Dacron graft.

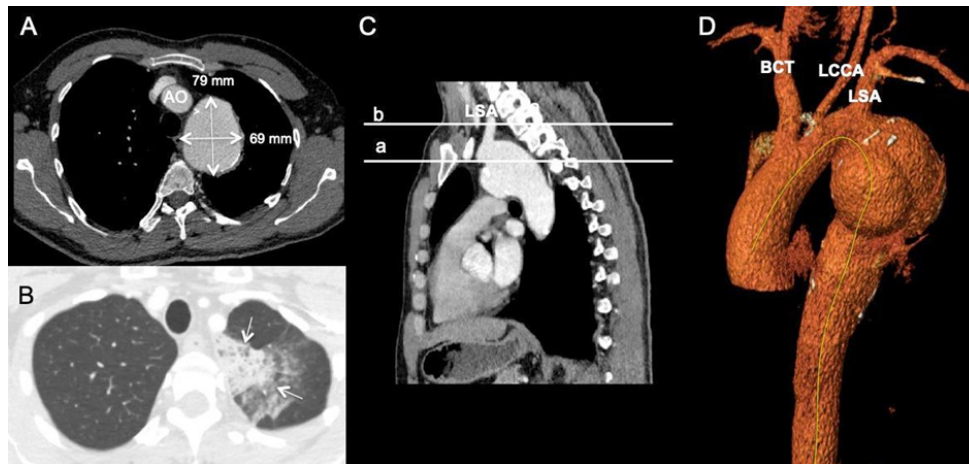


Figure 1: A: axial multidetector contrast-enhanced computed tomography (CT) examination of a bulky (79 x 69 mm) thoracic aortic pseudoaneurysm. B: lung window in the axial plan shows multiple pulmonary ground glass opacities in the apex (white arrows). C: coronal CT projection of a bulky (79 x 69 mm) thoracic aortic pseudoaneurysm starting just after the left subclavian artery (LSA). D: 3D-reconstruction of the CT.

Immediately after the bypass was established, a 170mm E-VITA thoracic 3D Jotec® endograft was expanded just distally to the brachiocephalic trunk in zone 1, using bilateral femoral approaches. The final completion angiogram showed a type Ib endoleak. The endoleak was immediately managed using a second E-VITA thoracic Jotec® endograft ending around the vertebral T9 level. A final selective angiogram demonstrated the complete exclusion of the aneurysm and the absence of endoleak. The brachio-cephalic trunk and the bypass were permeable (Figure 3). The total operative time was 221 minutes, clamping times were 7, 15 and 17 minutes for the right CCA, the left CCA and the vertebral

artery, respectively. Total blood loss was 200 ml. The patient was extubated immediately after surgery and transferred to the intensive care unit for 24 hrs. At post-operative day 2, the angioCT scan showed a permeable graft and supra-aortic debranching (Figure 4). Doppler pulse ultrasound examination confirmed good flow in the supra-aortic trunks with triphasic and symmetric flow (Q 500ml/min) in the subclavian arteries with no steal syndrome. The neurovascular examination of the arm was normal, with both radial and ulnar pulses well detectable. The patient was discharged from hospital and allowed to go home on post-operative day 5 without any complication or recurrence of hemoptysis.

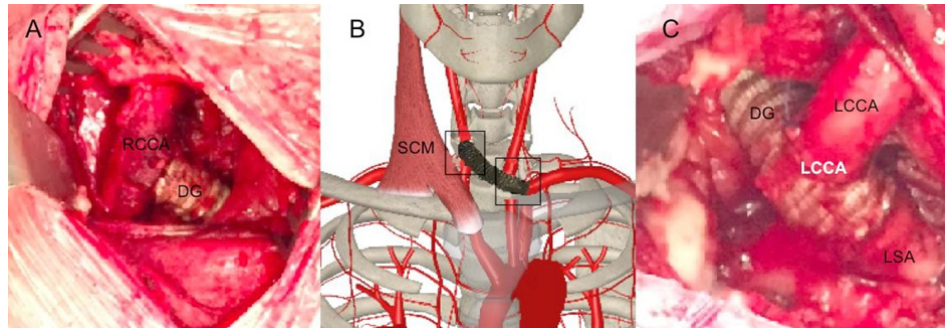


Figure 2: A: transverse incision over the anterior border of the right sternocleidomastoid (SCM), in the deep layer of the end-to-side anastomosis between the Dacron graft (DG) to the right common carotid artery (RCCA). B: Diagram of the bypass with the reimplantation of the left common carotid artery (LCCA). C: Left subclavian incision, in the deep layer of the two end-to-end anastomoses (DG to the left subclavian artery (LSA) and DG to the LCCA).

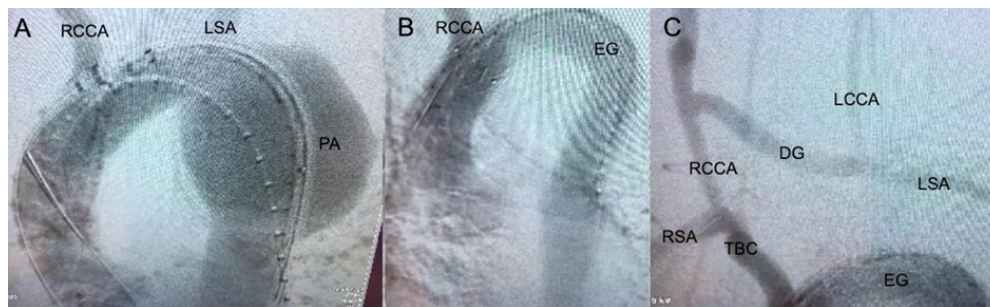


Figure 3: A: angiography before the endograft deployment (pseudoaneurysm (PA) starts just after the left subclavian artery (LSA) and spreads over approximately 6 cm). B: angiography after the endograft deployment (pseudoaneurysm is excluded), Endograft (EG). C: angiography of the permeable bypass graft (right common carotid artery (RCCA), Dacron graft (DG), left common carotid artery (LCCA), left subclavian artery (LSA)).

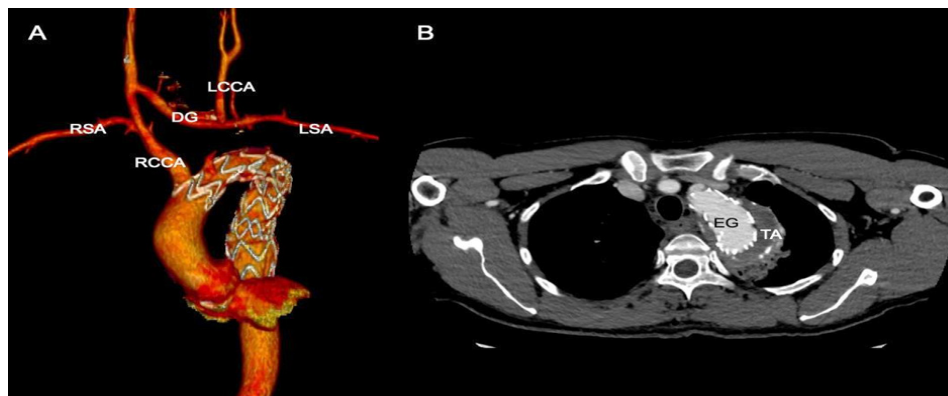


Figure 4: A: three-dimensional reconstruction computed tomography showing permeable endograft and bypass (right common carotid artery (RCCA), Dacron graft (DG), left common carotid artery (LCCA), left subclavian artery (LSA), right subclavian artery (RSA)). B: axial CT examination shows the thrombosed aneurysm (TA) and the patent endograft (EG).

Discussion

This case illustrates a rare occurrence of aorto-bronchial fistula (ABF) caused by a pseudoaneurysm many years after coarctation repair. Hemoptysis was the main clinical sign and diagnosis was confirmed by CT scan. Management was discussed in a multidisciplinary team including vascular, cardiac and thoracic surgeons. Kodolitsch and al [4] studied 25 pseudoaneurysms following aortic coarctation repair. In their cohort, none of them presented with ABF. Advanced age at coarctation repair and patch graft technique independently predicted local aneurysmal formation. Considering the large diameter of the pseudoaneurysm located in a field that had already been operated on twice, open surgery was deemed too dangerous, even considering the young age of the patient and the absence of medical comorbidities. This opinion was aligned with reports by other authors, which published relatively high rates of intra-operative deaths (15%) for similar indications, and 100% mortality with a conservative treatment and no surgery [1]. In another study, Mosquera and al [2] retrospectively reviewed 18 aorto-bronchial fistulas, of which five were secondary to aortic open repair. These authors studied two management techniques: open surgery and thoracic endovascular aortic repair (TEVAR) for patients in whom open surgery was not possible because of comorbidities. They assessed outcome predictors of aorto-bronchial fistulas and reported that with both surgical approaches, outcomes were mainly conditioned by the etiology of the fistula. These authors also reported that the worst prognosis was found among patients with fistulas caused by mycotic aneurysms and that the overall in-hospital mortality was 50 % (n=9), mostly caused by sepsis. Recently, endograft repairs became an alternative to open surgery with encouraging results even in patients suitable for open surgery. Bailey and al [3] studied 11 cases (mean age 69.8 years old, range: 30-89) of endovascular treatment of ABF. These authors reported a 91% technical success

rate during an average follow-up of 8.8 months. Endoleaks (9%) and infection (18%) were the most prevalent complications of this technique requiring long-term follow-up [4].

Conclusion

Pseudoaneurysm with ABF following aortic coarctation repair is a rare etiology of ABF with relatively non-specific clinical manifestations. Surgically, it is particularly challenging because it is frequently located at the aortic isthmus and it occurs in a young population that has already been operated at least once. Those characteristics require a durable and safe treatment in a patient with delicate ground and no alternative non-surgical options. No guidelines have been established as yet for the management of this rare complication. Our choice of a hybrid management technique was successful and outcomes at one month were positive.

References

1. Picichè, Marco, De Paulis R, Fabbri A, Chiariello L (2003) "Postoperative Aortic Fistulas into the Airways: Etiology, Pathogenesis, Presentation, Diagnosis, and Management." *The Annals of Thoracic Surgery* 75: 1998-2006.
2. Mosquera, Victor X, Milagros Marini, Pombo-Felipe F, Gómez-Martínez P, et al. (2014) "Predictors of Outcome and Different Management of Aortobronchial and Aorto-esophageal Fistulas." *The Journal of Thoracic and Cardiovascular Surgery* 148: 3020-26.e1-2.
3. Bailey, Charles J, Seth Force, Ross Milner, Karthikeshwar Kasirajan, et al. (2011) "Thoracic Endovascular Repair as a Safe Management Strategy for Aortobronchial Fistulas." *Journal of Vascular Surgery* 53: 1202-1209.
4. Von Kodolitsch Y, Aydin MA, Koschyk DH, Loose R, Schalwat I, et al. (2002) "Predictors of Aneurysmal Formation after Surgical Correction of Aortic Coarctation." *J Am Coll Cardiol*. 39: 617-624.