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CASE SERIES
Other

Thoracic endovascular aortic repair for aortobronchial fistula: a case series

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Background Aortobronchial fistula (ABF) formation following the rupture of thoracic pseudoaneurysm is a rare clinical entity. Its aetiology includes atherosclerosis, infections, trauma, post-surgery, and post-endovascular aortic repair. The clinical presentation of ABF includes intermittent or massive haemoptysis, acute respiratory distress, hypotension, and even death. These patients require an emergency aortic intervention to stop active haemorrhage. Thoracic endovascular aortic repair (TEVAR) is a less invasive, safe, and effective treatment compared to conventional open surgical repair

Case summary We hereby report three cases of ruptured descending thoracic aortic pseudoaneurysms resulting in a fistula formation. The first two cases had tuberculosis as their underlying aetiology, while the third case was the result of previous open post-aortic surgery. All patients presented with massive haemoptysis and were successfully treated by emergency TEVAR and had favourable outcomes.

Discussion Thoracic endovascular aortic repair is a rapid, less invasive, and effective treatment for emergency management of ABF. It has more than 85% technical success rates in the reported literature. We had procedural success in all three cases. The short and midterm outcome of ABF following TEVAR is favourable and encouraging.

Keywords Aortobronchial fistula • Massive haemoptysis • Thoracic endovascular aortic repair • Graft stent • Thoracic pseudoaneurysm • Tuberculosis • Case series

Learning points

- Aortobronchial fistula is a rare disease and always an emergency.
- Tuberculosis is one of the less common aetiologies.
- Conventional open surgical repair is associated with high morbidity and mortality.
- Thoracic endovascular aortic repair is a less invasive treatment modality with good procedural success and with good short and middle term outcomes.

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Introduction

Aortobronchial fistula (ABF) formation following the rupture of aortic pseudoaneurysm is a rare and invariably fatal clinical entity. Its aetiologies include atherosclerosis, infections including tuberculosis, trauma, and post-interventions such as aortic surgery, coarctation repair, endovascular aortic treatment, and esophagectomy.[1–4] Tuberculosis related arteritis and pseudoaneurysm is a rare cause of ABF.[5] Massive haemoptysis and exsanguination following its rupture in lung parenchyma can result in acute respiratory distress, hypotension, and death if an emergency intervention is not performed. An endovascular stent-graft repair of the ruptured aorta can be an alternative to conventional surgery, which otherwise carries high morbidity and mortality.[2,6–8] We hereby report three cases of ABF following aortic pseudoaneurysm rupture, which were successfully treated with thoracic endovascular aortic repair (TEVAR).

Timeline

Patient 1	
4 months before presentation2 months before presentation	A 30-year-old male had back painHad fever and cough with expectorationDiagnosed with tuberculosis, by sputum microscopy and started on anti-tubercular therapy (ATT)
Day 0 (1 month after ATT)	Presented with haemoptysis. A computed tomography (CT) angiography showed 25 mm × 15 mm of pseudoaneurysm from descending thoracic aorta (DTA) with surrounding haematoma and diagnosed as tubercular pseudoaneurysm with aortobronchial fistula. Underwent successful thoracic endovascular repair (TEVAR) with Valiant 26 mm × 26 mm × 100 mm thoracic graft stent
4 months of follow-up	Asymptomatic. Repeat CT showed no endoleak and regression of pseudoaneurysm
Fifth month post-procedure	Presented with rapidly progressive dyspnoea. Investigations were inconclusive for the relapse of tuberculosis. He had a fulminant course during the hospital stay and succumbed to respiratory failure
Patient 2	
5 months before presentation	A 26-year-old female diagnosed with brain tuberculoma and started on ATT
Day 0	Presented with haemoptysis. CT angiography showed two pseudoaneurysms of size 16 mm × 10 mm and 3.4 mm × 7 mm with surrounding haematoma
	Underwent successful TEVAR with Valiant 22 mm × 22 mm × 100 mm graft stent
At 8 months follow-up	Repeat CT showed no endoleak with the regression of pseudoaneurysms
At 5 years of follow-up	Doing well with no recurrence of symptoms
Patient 3	
3 months before presentation	A 24-year-old male had a blunt chest injury following a road traffic accident, resulting in transection of DTA. Underwent emergency open aortic repair with Dacron tube graft
Day 0	Presented with haemoptysis after 3 months. CT angiography revealed an 11 mm × 17 mm pseudoaneurysm of DTA along the suture line with surrounding haematoma
	Underwent successful TEVAR with Valiant 32 mm × 28 mm × 150 mm stent-graft
At 3 months follow-up	Repeat CT showed no endoleak with the regression of pseudoaneurysm
At 3 years of follow-up	Doing well with no recurrence of symptoms

Case presentation

Case 1

A 30-year-old male presented with low back pain for the last 4 months, and fever with mucoid expectoration for last 2 months. His clinical examination was unremarkable. A chest X-ray was normal without any lung parenchymal lesion. Sputum examination confirmed the presence of acid-fast bacilli, for which he was started on anti-tubercular therapy (ATT). After a month, he presented with three bouts of massive haemoptysis, for which two units of blood transfusion were given. A computed tomography (CT) chest revealed multiple, enlarged perihilar, and mediastinal lymph nodes, without any significant lung parenchymal disease. A contrast CT angiography of aorta revealed a 25 mm × 15 mm pseudoaneurysm arising from the lower part of descending thoracic aorta (DTA), with peri-aneurysmal haematoma (*Figure 1A*). The proximal DTA diameter was 20 mm, while right and left common femoral arterial diameter was 7.9 and 7.4 mm, respectively. He underwent an emergency TEVAR, using a 26 mm × 26 mm × 100 mm thoracic stent-graft (Valiant Captivia Thoracic Stent, Medtronic Inc., Minneapolis, MN, USA) through right femoral access, under general anaesthesia (*Figure 1B and C*). The

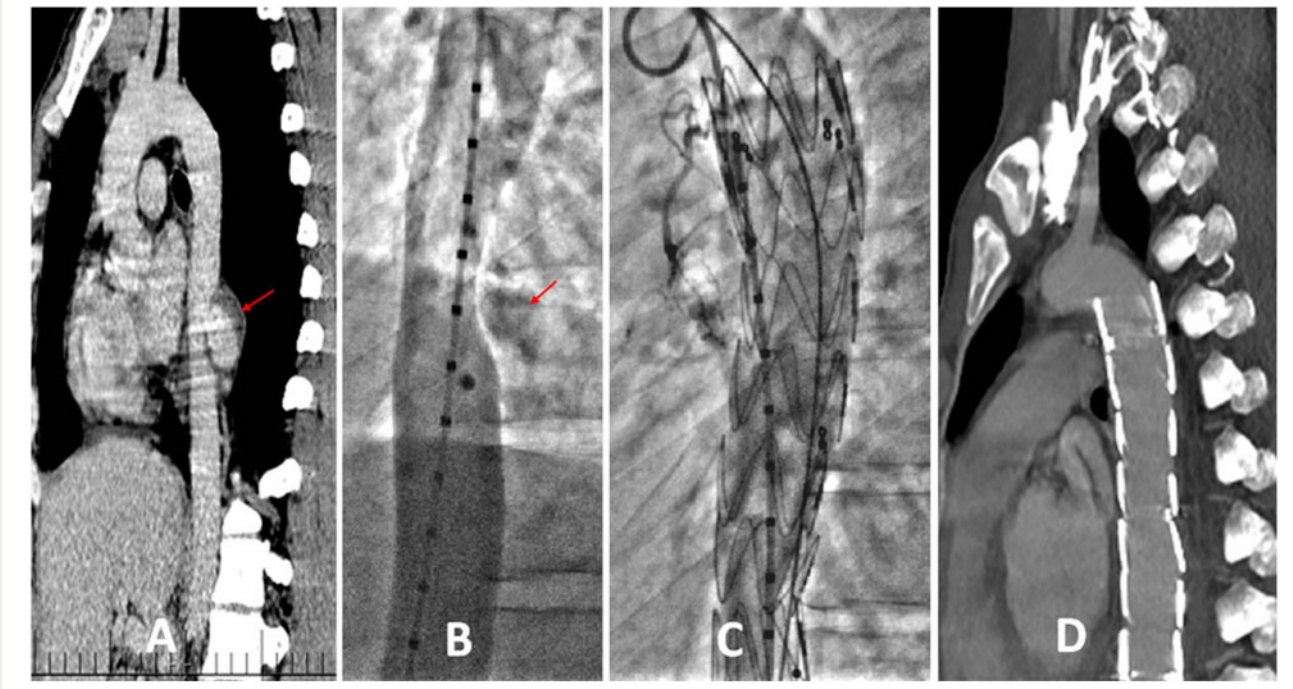

Figure 1 A computed tomography in sagittal, oblique reformatted view showed a 25 mm × 15 mm pseudoaneurysm with surrounding haematoma arising from the lower part of descending thoracic aorta (*A*). Contrast angiogram showed the filling of the pseudoaneurysm (*B*), which was excluded by a stent-graft (*C*). A repeat computed tomography at 4 months of follow-up showed patent aortic lumen, regression of pseudoaneurysm with no endoleak (*D*).

surgically explored right femoral artery was repaired following the completion of the procedure. Post-procedure, he remained asymptomatic for the next 4 months of follow-up, on ATT. Later, he was again admitted with recurrent cough, streaky haemoptysis, and worsening dyspnoea. His workup for tuberculosis reactivation was inconclusive. A repeat CT scan showed patent stent-graft, no endoleak, and complete regression of the pseudoaneurysm (*Figure 1D*). However, he had a fulminant course and died of aspiration pneumonitis and respiratory failure.>

Case 2

A 26-year-old female who was on ATT from the last 5 months for brain tuberculoma, presented with four bouts of massive haemoptysis to the emergency room. She received three units of blood transfusion for excessive blood loss. Her clinical examination including the central nervous system was unremarkable. The chest X-ray showed a parenchymal fibrotic/consolidative patch in the left mid-zone abutting the DTA. A CT angiography of aorta revealed two small saccular pseudoaneurysm of size 16 mm × 10 mm and 3.4 mm × 7 mm, arising from the upper part of DTA with surrounding haematoma (*Figure 2A*) and lung parenchymal changes. There was no significant hilar or mediastinal lymphadenopathy. The proximal DTA diameter was 17–18 mm, while the diameter of the right and left common femoral artery was 5.5 and 4.9 mm, respectively. She was considered for emergency TEVAR, under general anaesthesia. As the size of bilateral femoral arteries were too small to pass a 22 Fr stent-graft, an open surgical Dacron conduit was anastomosed to the right common iliac artery to facilitate the delivery of stent-graft at DTA. The procedural details about surgical iliac conduits to deliver large-sized endograft aortic devices have been described in details by Peterson and Matsumura.[9] A 22 mm × 22 mm × 100 mm thoracic stent-graft (Valiant Captivia Thoracic Stent, Medtronic Inc.) (*Figure 2B and C*) was deployed covering the DTA pseudoaneurysm. After completion of the endovascular procedure, the iliac conduit was amputated at the base, leaving behind a small stump attached to the iliac artery. Post-procedure, she had an uneventful recovery and discharged on ATT of a total 9 months duration. A repeat CT angiography at 8 months of follow-up showed patent stent-graft, no endoleak, and complete regression of the pseudoaneurysm (*Figure 2 D*). She remained asymptomatic during the next 5 years of follow-up.

Case 3

A 24-year-old male had acute transection of DTA in a road traffic accident, for which he underwent successful emergency open surgical repair using a Dacron tube graft. Three months later, he presented with one bout of massive haemoptysis, requiring two units of blood transfusion. His clinical examination was unremarkable. A CT angiography showed an 11 mm × 17 mm pseudoaneurysm with surrounding haematoma at the surgical anastomotic site of the DTA (*Figure 3A*). The diameter of DTA at the origin of the left subclavian artery was 26–27 mm. The diameter of the right and left common femoral artery was 7.25 and 8.25 mm, respectively. He underwent emergency TEVAR, using a 32 mm × 28 mm × 150 mm thoracic stent-graft (Valiant Captivia Thoracic Stent, Medtronic Inc.) through

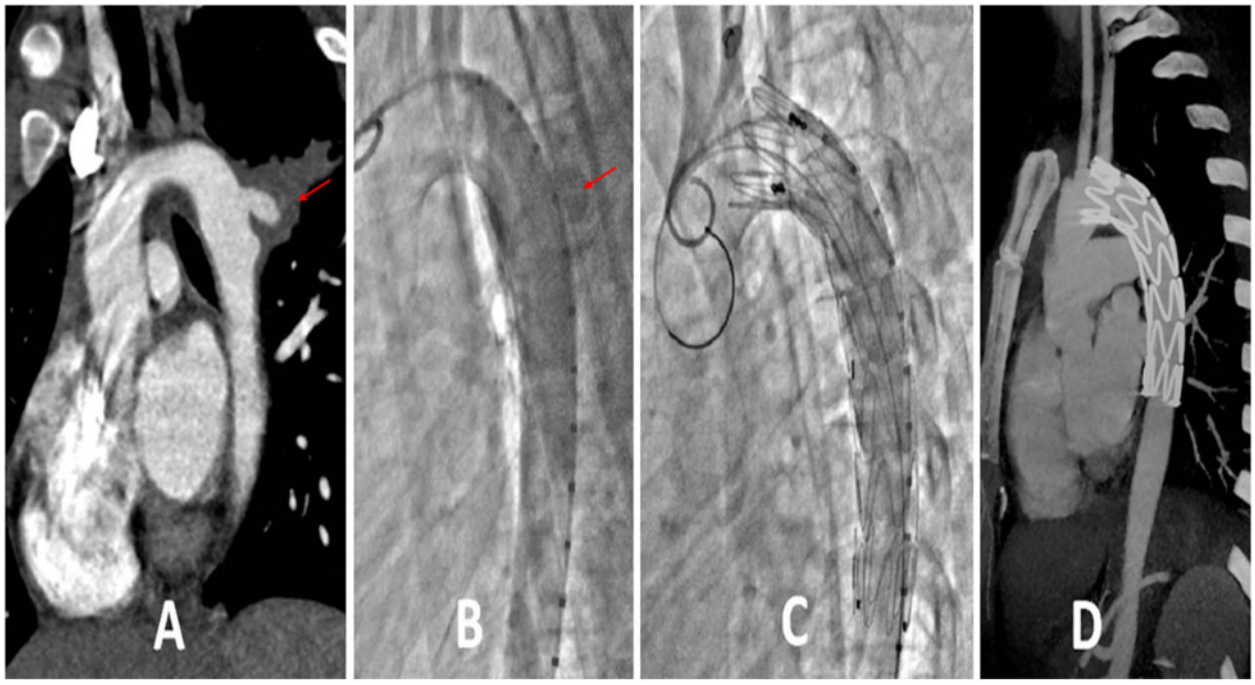

Figure 2 A computed tomography angiogram of the thoracic aorta in oblique, sagittal reformatted view showed two small saccular pseudoaneurysms arising from the upper part of descending thoracic aorta with surrounding haematoma and parenchymal consolidation (*A*). Contrast angiogram showed the filling of pseudoaneurysms (*B*), which was excluded by a stent-graft (*C*). A repeat computed tomography at 8 months of follow-up showed patent aortic lumen and regression of pseudoaneurysm with no endoleak (*D*).

the left femoral access, under general anaesthesia (*Figure 3B and C*). The left subclavian artery was covered with the stent-graft to achieve an adequate seal. The surgically explored left femoral artery was repaired following the completion of the procedure. A repeat CT angiography at 3 months showed patent stent-graft, no endoleak, and complete regression of pseudoaneurysm (*Figure 3D*). He remained asymptomatic during the next 3 years of follow-up.

Discussion

We found tuberculosis and previous aortic surgery as the aetiology for aortic pseudoaneurysm in three patients. Various authors have reported atherosclerosis, previous aortic surgery, and mycotic aneurysm as the common aetiology of ABF,[1,2,6,7] while tuberculosis is a rare cause for it.[5] Previous aortic surgery is a common risk factor for pseudoaneurysm formation. It usually occurs at the suture line or at the cannulation site due to iatrogenic defects in the vessel wall and subsequent poor healing.[10] Tubercular aortitis is a rare aetiology for ABF. Tubercular aortic involvement is either by spread from adjacent periaortic structures such as lymph nodes, pleura, lung, vertebra,[11–13] or directly from the blood circulation. Direct bacterial seeding of luminal surface and aortic wall transmission via lymphatics and vasa vasorum are the pathological mechanisms for its haematogenous spread.[13] Case 1 had adjacent infected lymph nodes, while Case 2 had possible systemic bacteraemia or contiguous spread from adjacent lung as the cause for tubercular aortic pseudoaneurysm.

The clinical presentation of ABF includes intermittent or massive haemoptysis, blood loss, acute respiratory distress, hypotension, and even death.[1,2] A high clinical suspicion is required for its diagnosis in a patient presenting with massive haemoptysis, DTA disease, and with/without parenchymal lung disease.[2] The CT angiography may not directly demonstrate fistula, but certain subtle findings such as the presence of air in thrombus, periaortic blood/thrombus collection, small pseudoaneurysm, bronchial wall thickening, and lung consolidation adjacent to aneurysm can suggest the presence of ABF.[1,2] Bronchoscopy may be helpful but has a risk of dislodging the thrombus leading to massive haemoptysis and death.[1,2] All of our three cases had adjacent haematoma at the site of pseudoaneurysm along with massive haemoptysis, which was suggestive of ABF.

As the patient remains haemodynamically unstable because of excessive blood loss, emergency intervention is required in such a situation. The conventional open surgical repair requires thoracotomy, prosthetic bypass, or homograft reconstruction of the DTA along with bronchial/lung tissue resection, which carries high mortality ranging from 16% to 24%.[8,14,15] Picichè *et al.*[14] in a systematic analysis of 50 patients of ABF reported a 16% mortality rate following open surgical repair. MacIntosh *et al.*[8] in a review of 34 patients of open surgical repair reported a 24% 30-day mortality. Eren *et al.*[15] in another series of 10 patients reported a mortality of 20% during the perioperative period. The high morbidity and mortality associated with the open repair are due to the emergent situation of the disease, the complex nature of the surgery, associated risk of graft infection

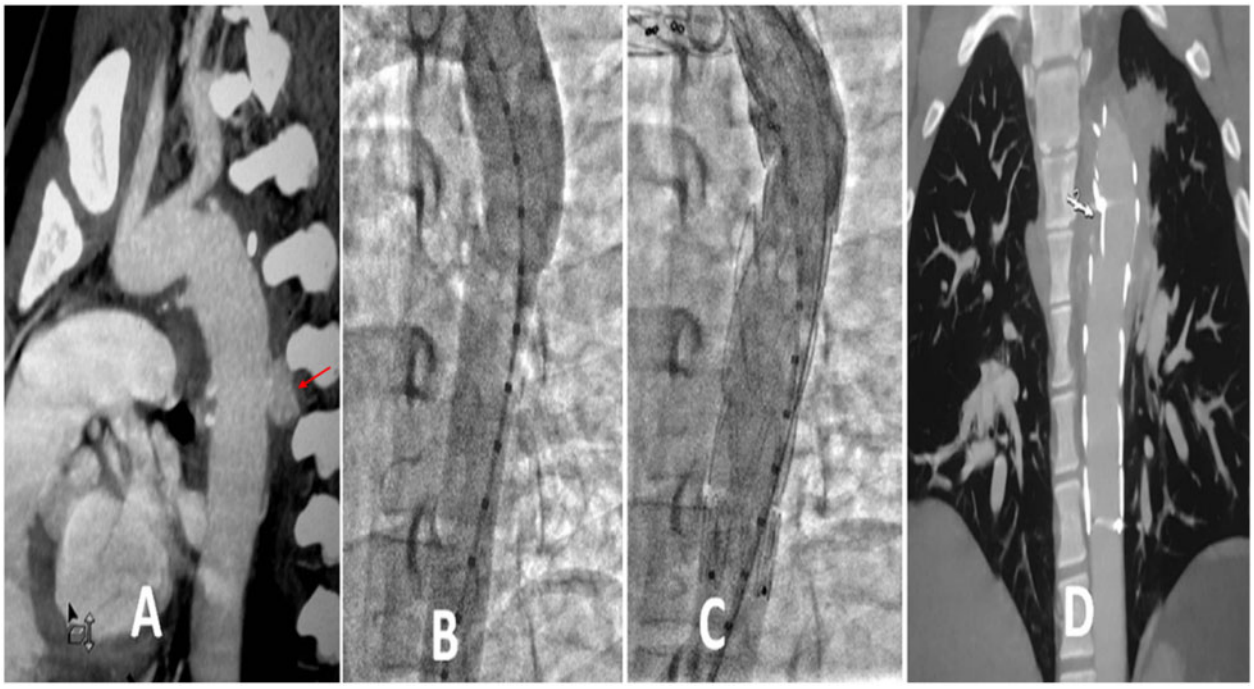

Figure 3 A computed tomography angiogram in sagittal, reformatted view showed an 11 mm × 17 mm pseudoaneurysm at the mid-part of descending thoracic aorta, at surgical anastomotic site (*A*). A contrast angiogram could delineate the surgical anastomotic site with a pseudoaneurysm (*B*), which was excluded by a stent-graft (*C*). A repeat computed tomography at 3 months of follow-up showed patent descending thoracic aorta lumen and regression of pseudoaneurysm with no endoleak (*D*).

and sepsis, and the need for a recurrent procedure in the postoperative period.[1,2,8,14,15]

Thoracic endovascular aortic repair has emerged as an alternative, less invasive intervention compared to standard open surgical repair in such patients.[1,2,6,7,16] Canaud *et al.*[1] in a systematic review of 134 ABF patients subjected to TEVAR reported a 93% technical success rate. The 30-day mortality was 5.9%, while 17 months aortic and all-cause mortality was 14.3% and 21.4%, respectively.[1] Kawaharada *et al.*[2] in a case series of 26 patients of ABF treated with TEVAR reported a procedural success rate of 85% and 30-day mortality of 15%. In an Italian survey of 25 patients of both aortobronchial and aorto-oesophageal fistulas, TEVAR was associated with a 30-day mortality of 28%.[6] The higher mortality was possibly because of the inclusion of aorto-oesophageal fistula, which had a relatively worse outcome.[6] Thoracic endovascular aortic repair is again a preferred first line of treatment for mycotic pseudoaneurysm in high-risk patients.[17] These patients require long-term antibiotic coverage, have a risk of graft infection, and may require additional open surgery to explant the infected stent-graft.[1,2,17] There are only a few published case reports of the tubercular pseudoaneurysm with ABF, which were treated with TEVAR.[18–20] We had technical success of TEVAR in all three patients and none had mortality within 30 days. Certain complications following TEVAR such as graft infection, endoleak, recurrence of haemoptysis/fistula formation, sepsis, multi-organ failure, and need for surgical explantation of infected stent-graft have been reported.[1,2] Canaud *et al.*[1] in a systemic review found an 11% recurrence rate of fistula and 3.5% surgical conversion rate after TEVAR.[1] The risk factors for recurrence were Type I endoleak, stent-graft erosion causing ischaemic necrosis of surrounding bronchus, renal dysfunction, and respiratory failure.[1,2,6,21] The preferred intervention for ABF in the last two decades is TEVAR instead of primary open surgical repair. Open surgery is mostly a secondary intervention in post-TEVAR cases to manage complications such as fistula recurrence, stent-graft infection, Type 1 endoleak, and aortic injury.[1,21,22] Those with infected stent-graft require open surgery for stent removal and aortic revascularization using various techniques.[23] All the three patients did not experience any such complications. The recommended duration of the ATT for tubercular pseudoaneurysm is 6–9 months.[24] Our Case 2 recovered well after 9 months of ATT, while Case 1 died prematurely after receiving 5 months of ATT. None of the patients had any clinical or radiological evidence of stent-graft infection at follow-up. Cases 2 and 3 had asymptomatic follow-up of 3 and 5 years, respectively.

In conclusion, we hereby report three cases of ABF, which were successfully treated with TEVAR and had favourable outcomes. One case of tuberculosis pseudoaneurysm died unrelated to the intervention at 4 months of follow-up, while the other two patients had favourable long-term outcomes. Thoracic endovascular aortic repair is an alternative, less invasive treatment with a lower mortality rate, compared to the open surgical repair in selected patients of ABF.

Lead author biography

Prof Dr Rajesh Vijayvergiya, MD, DM, FACC, FSCAI, FISES, is working as Director, Catheterization lab at Post Graduate Institute of Medical Education and Research (PGIMER), Chandigarh. His area of interest is percutaneous coronary and peripheral arterial interventions. He has published 140 papers in various national and international journals, 12 chapters in various books and is a member of the editorial board of 11 national and international journals. He is the national coordinator from India for the European Association of Percutaneous Cardiovascular Interventions (EAPCI) educational programme.

Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as Supplementary data.

Consent: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patients in line with COPE guidance.

Conflict of interest: none declared.

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