Late Aortic Rupture Due to Stent Margin Pseudoaneurysm Formation Complicating Endovascular Stent Graft Repair of a Thoracic Aortic Mycotic Aneurysm

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Acute rupture of a thoracic mycotic aneurysm is a surgical emergency that requires urgent treatment to prevent further bleeding. The results of surgical repair are limited by high mortality and paraplegia rates. Endovascular repair with stent graft deployment has been reported as an alternative to high-risk surgery. We present the case of a patient with a ruptured mycotic aneurysm of the thoracic aorta complicated by an aorto-oesophageal fistula. Endovascular stent graft deployment was successful at controlling bleeding. Oesophagectomy was required for a persistent leak from the oesophagus. The patient remained stable for 13 months after the index procedure and died from aortic rupture. This was related to the development of a pseudoaneurysm of the aortic arch immediately proximal to the stent margin. This complication may result from mechanical factors related to the presence of a stent graft.

Keywords: Stent graft; Endovascular repair; Mycotic aortic aneurysm; Aorto-oesophageal fistula; Late rupture.

Introduction

Aortic mycotic aneurysm is a fulminant infectious disease that can progress to aortic rupture and death. Conventional treatment consists of surgical repair which requires a lateral thoractomy, aortic cross clamping and circulatory bypass. Surgical options include the in situ placement of an aortic allograft or the creation of an extra-anatomic bypass with surgical resection of the infected material.1,2 The results of surgery are limited by a perioperative mortality exceeding 50% in patients with aortic rupture and a paraplegia risk of 7-17%.3,4 Patients are often not suitable for surgery due to coexisting medical problems. Endoluminal stent grafts have been used as an alternative to surgery for the treatment of patients with acute rupture of the descending thoracic aorta.5 This technique has the advantages of a minimally invasive approach, immediate control of bleeding and a relatively short procedural time. We describe the case of a patient with an aorto-oesophageal fistula secondary to a ruptured mycotic thoracic aneurysm treated with endovascular stent graft deployment.

Case Report

A 55 year old lady was admitted acutely with back pain. She subsequently developed haematemesis and meleana. Endoscopy showed a pulsatile bleeding point in the mid oesophagus with the appearances of an aorto-oesophageal fistula. A computed tomography (CT) scan of the thorax showed a pseudoaneurysm of the thoracic aorta with periaortic haematoma and a left haemothorax. The patient was transferred to our regional cardiothoracic centre for further management.

Past medical history included end stage renal failure due to bilateral nephrectomy for chronic pyelonephritis 20 years previously. A cadaveric renal transplant had recently failed requiring the re-establishment of haemodialysis. The patient had been on antibiotics for the last three weeks due to an infected right internal
jugular dialysis line and *Staphylococcus aureus* septicemia. The patient was considered too unwell to undergo open surgical repair and an endovascular aortic stent graft procedure was performed in March 2002. This was carried out in a cardiac angiography suite under local anaesthesia. The left common femoral artery was exposed to allow the delivery of the stent graft. Systemic anticoagulation was achieved with an intravenous injection of heparin after vascular access had been secured. A 34 mm by 10 cm Talent stent graft (Medtronic, Sunrise, USA) was deployed at the site of the thoracic aortic aneurysm (Fig. 1). This self-expandable endovascular prosthesis is constructed out of circumferential nitinol stent rings arranged as a tube and covered on the outer surface with a Dacron graft. The individual stent rings are secured to the graft material with oversewn sutures. The stent rings are not connected to one another and there are segments of unsupported graft interposed between the stents. Two additional longitudinal wires are used to provide stabilization and prevent compression. The ends of the stent graft consist of bare springs that provide support for effective anchoring to the vessel wall whilst allowing side branch access. The latest design of the Talent stent is available with fabric covering along the entire length or with bare metal springs at the proximal end only. Post implantation aortogram showed no endovascular leak and the procedure was considered a success. A chest drain was inserted to drain the infected left haemothorax and intravenous antibiotics were continued. A contrast CT scan of the thorax on day 3-post procedure showed leakage of contrast into the thoracic aortic aneurysm. An aortogram showed persistent perfusion of the aneurysm sac. This was thought to represent either a type I or type III endoleak. Balloon moulding of the stent did not abolish the leak and two further Talent stent grafts were deployed, with overlap proximal and distal to the original stent (Fig. 2). This resulted in triple overlapping layers of stent over the central portion of the aortic aneurysm. The proximal stent graft extended into the descending portion of the aortic arch, distal to the origin of the left subclavian artery. No further endoleak was seen on repeat aortogram. The patient’s recovery was complicated by mediastinal sepsis caused by a persistent leak from the oesophagus. An oesophagectomy and left decortication was performed on day 22-post stent implantation. The delay in surgery was due to our initial attempts to manage the patient conservatively as she was considered high-risk for surgery due to her co-morbidity and frail condition. The necrotic portion of the oesophagus was excised and the distal segment closed. The proximal section of the oesophagus was brought to the anterior chest wall as

Fig. 1. Talent Stent Graft (Medtronic, Sunrise, USA).

Fig. 2. Aortogram in the lateral projection after deployment of the Talent stent grafts.
a stoma. A feeding jejunostomy was placed in the proximal jejunum. No active bleeding from the thoracic aorta was noted at the time of surgery. Postoperative recovery was complicated by septicemia and nutritional problems. Following a prolonged hospital stay the patient was discharged home 6 months after the index procedure off antibiotics. A CT scan at this stage showed no evidence of endoleak. Oesophageal reconstruction was performed in February 2003 following which normal eating was re-established.

The patient remained stable until 13 months after the original procedure when her condition deteriorated. A repeat CT scan showed the presence of a pseudoaneurysm at the interface between the bare stent and the unstented aorta, which was not present on previous studies (Figs. 3 and 4). The patient died before any further intervention could be performed. A post mortem examination showed that the cause of death was rupture of the thoracic aorta. The entire thoracic aorta, at a point just distal to the origin of the left subclavian aorta was aneurysmal and friable. A 9 mm defect was noted in the descending thoracic aorta just proximal to the stented segment. The endovascular stent grafts were incorporated into the wall of the aorta with a patent lumen. Tissue samples from the site of aortic rupture showed no evidence of active infection on both histology and microbiology.

Discussion

A mycotic aneurysm is defined as a localised irreversible dilation of an artery to at least one and one half times its normal diameter due to destruction of the vessel wall by an infection. Mycotic aortic aneurysms are rare accounting for 0.9% of all aortic aneurysms undergoing reconstructive surgery. Sir William Osler first coined the term “mycotic aneurysm” to denote an appearance like “fresh fungal vegetation”. Mycotic aneurysms can occur following bacterial infection of a previously normal arterial wall or through secondary infection of a pre-existing aneurysm. Risk factors for mycotic aneurysms are infective endocarditis, septicemia, intravenous drug use, immunosuppression and pre-existing aneurysm. The commonest infecting organisms are Staphylococcus aureus, Salmonella and Streptococcus species. The first clinical manifestation is often related to aneurysm rupture that can involve the pleural cavity, oesophagus, trachea or bronchus. Rupture into the oesophagus is associated with the formation of an aorto-oesophageal fistula and catastrophic gastrointestinal bleeding. The classic presentation consists of midthoracic chest pain, sentinel bleeding followed by fatal arterial bleeding. More than 50% of patients die within 6 hours after the first arterial bleed. Survival depends on early diagnosis and treatment to prevent further bleeding.
Late Aortic Rupture Due to Stent Margin Pseudoaneurysm

Stent graft repair of ruptured thoracic aortic aneurysms is emerging as an alternative to high-risk surgery. This technique allows for immediate patching of the leak and can be used as a stand-alone treatment or for stabilisation prior to surgery. Endovascular treatment is associated with a lower in hospital mortality and reduced risk of paraplegia compared to surgery. The presence of aortic infection leading to mycotic aneurysm formation increases the risk of endovascular therapy. An inflamed and degenerative aortic wall may be so fragile that adequate anchoring of a stent graft may not be possible. This would result in persistent flow into the aneurysm even after stent graft deployment. The deployment of an endovascular prosthesis at the site of active infection would increase the risk of stent graft infection. Infections involving endovascular stents have been reported and these require removal of the prosthesis and surgical reconstruction.

Semba et al. successfully treated three patients with mycotic aneurysms of the thoracic aorta, including one with a ruptured aneurysm using stent graft placement and adjunctive antibiotics. There were no cases of persistent sepsis during a follow up period of 4-25 months. Gonzalez-Fajardo et al. reported a case of aorto-oesophageal fistula complicating mycotic aortic aneurysm rupture treated with endovascular stent implantation. Although gastrointestinal bleeding was successfully controlled with stenting, the patient died 7 days post procedure due to uncontrolled sepsis and aortic rupture. Van Doorn et al. described a case where endovascular stent implantation was used to stabilise a patient with upper gastrointestinal bleeding from an aorto-oesophageal fistula associated with a mycotic aortic aneurysm. The patient underwent oesophagectomy, mediastinal irrigation and debridement three days after stenting and was doing well 2 years post procedure.

This case report illustrates several important technical issues in the endovascular treatment of aorto-oesophageal fistula complicating mycotic aneurysm rupture. Leakage from the oesophagus may persist even after endoluminal sealing of the aortic aneurysm.

This would result in mediastinitis and increased risk of stent graft infection. We would recommend that endovascular stent graft placement be used as a primary procedure to control bleeding. In patients who are suitable for surgery, oesophagectomy and mediastinal drainage should be performed due to the risk of continued leakage from the oesophagus. Previous reports have shown that in patients with acute thoracic aortic rupture, rates of repeat intervention and late complications are higher with endovascular intervention compared to surgical repair. Doss et al. reported late aortic rupture in 6.2% of patients at four years after endovascular repair of acute thoracic aortic rupture. Demers et al. reported late aortic rupture in 9% of the patients at five years following endovascular repair of descending thoracic aortic aneurysms. The commonest late complication following endovascular repair of thoracic aortic aneurysms is type I endoleak. This occurs due to loss of fixation at the proximal or distal attachment sites, and may be related to device migration or aneurysm neck expansion. Endoleaks result in recurrent pressurisation of the aneurysmal sac that can progress to aortic rupture. Retrograde aortic dissection due to a tear in the aortic wall proximal to the stent graft has been reported to occur following endovascular thoracic aortic repair. The aortic tear is often caused by wire and sheath handling or balloon dilatation during the endovascular procedure. This complication presents days or weeks after stent graft implantation.

Our case illustrates another potential cause of late aortic rupture after endovascular stent deployment. Mechanical strain on the aorta from the bare metal springs of the endovascular stent with a hinge point between the rigid stented aorta and the native aorta may result in pseudoaneurysm formation. This may have been caused by disease progression although there was no evidence of active infection on histology or microbiology.

Our normal policy for surveillance after endovascular repair of a mycotic aneurysm is imaging with either computed tomography scan or magnetic resonance imaging every 3 months for the first year after discharge. For the second year imaging is performed 6 monthly and then once a year if the patient is stable. This case highlights that the reporting radiologist did not record the junctional pseudo-aneurysm that resulted in fatal aortic rupture.

Improvement in stent graft deployment to ensure coverage of the entire aneurysmal segment of the thoracic aorta and the avoidance of creating a proximal hinge point may reduce the risk of developing a junction pseudoaneurysm. More flexible stent grafts with fabric covering along the entire length may also reduce local forces on the aortic wall. This serious complication should be actively looked for during follow-up and aggressively treated when found.

References


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