SHORT REPORT

Aorto-enteric Fistula 15 Years After Uncomplicated Endovascular Aortic Repair with Unforeseen Onset of Endocarditis

M.M.K. Kadhim a,*, J.B.G. Rasmussen b, J.P. Eiberg a,c,d

- ^a Department of Vascular Surgery, Rigshospitalet, Copenhagen, Denmark
- ^b Department of Interventional Radiology, Rigshospitalet, Copenhagen, Denmark
- ^c Copenhagen Academy for Medical Education and Simulation, Rigshospitalet, Denmark
- ^d University of Copenhagen, Copenhagen, Denmark

Introduction: Aorto-enteric fistula after endovascular aortic repair is an exceedingly rare but serious condition. Report: A rare case of a fistula between the excluded aortic sac and the transverse colon 15 years after endovascular aortic repair is described. Onset was endocarditis without gastrointestinal haemorrhage, migration, endoleak, or aortic sac dilatation. The patient was successfully treated by fistula excision, debridement and broad spectrum antibiotic treatment without endograft explantation.

Discussion: Aorto-enteric fistula can emerge after endovascular repair with an unforeseen onset such as endocarditis, which in this case probably occurred as metastatic sepsis from endograft infection.

© 2016 The Authors. Published by Elsevier Ltd on behalf of European Society for Vascular Surgery. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Article history: Received 17 November 2015, Revised 14 March 2016, Accepted 15 March 2016,

Keywords: Aorto-enteric fistula, Endocarditis, Endovascular graft, EVAR

INTRODUCTION

A rare case of aorto-enteric fistula (AEF) formation between the aortic sac and the transverse colon with a most unusual and late onset is described.

CASE REPORT

A 66 year old man was admitted with confusion and fever (maximum temperature 38.5 $^{\circ}\text{C})$ for 2 days.

The patient had a history of hypercholesterolaemia, previous coronary artery bypass graft, left ventricular ejection fraction (LVEF) 20%, moderate mitral insufficiency, moderate aortic stenosis, cardiac re-synchronisation therapy device and prostatic hypertrophy.

Fifteen years prior to admission an EVAR, with implantation of a Zenith Tri-Fab stent graft (Cook Europe, Bjæverskov, Denmark), for a 7 cm infrarenal abdominal aortic aneurysm (AAA) had been performed. The patient followed the usual EVAR surveillance protocol, including yearly computed tomography angiography, abdominal ultrasound, and plain abdominal radiography, without any signs of migration, fracture, endoleak, or change in aneurysm sac size.

On admission, the patient had increased inflammatory parameters (white blood cell count: 14,700/µL; C-reactive

* Corresponding author.

E-mail address: muradkopurlu@hotmail.com (M.M.K. Kadhim).

2405-6553/© 2016 The Authors. Published by Elsevier Ltd on behalf of European Society for Vascular Surgery. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/). http://dx.doi.org/10.1016/j.ejvssr.2016.03.004

protein: 59 mg/L) and empirical antibiotic treatment with cefuroxime 1500 mg \times 3 intravenously was started. Within 2 days a trans-esophageal echocardiogram visualised vegetations on the mitral valve and as blood cultures revealed non-haemolytic *Streptococcus*, antibiotic treatment was changed to benzylpenicillin and rifampicin, according to the national guidelines regarding streptococcal endocarditis.

In pursuit of the infectious focus a computed tomography (CT) scan showed air bubbles in the otherwise stable aneurysm sac (Fig. 1), and the patient was transferred to the authors' department on suspicion of graft infection.

Extended bacteriological examination, including circulating immune complexes, polymerase chain reaction, and additional blood cultures, revealed *Enterococcus faecium* bacteraemia and antibiotic treatment was further intensified to include meropenem, rifampicin, and ciprofloxacin.

A positron emission tomography—CT scan was performed, demonstrating increased focal uptake from the stent graft and aneurysm sac, and a possible communication between the aneurysm sac and the transverse colon (Fig. 2).

After 14 days of antibiotic treatment, the patient underwent laparotomy and a para-prosthetic fistula between the transverse colon and the aneurysm sac was confirmed. The sac was filled with purulent material although the stent graft was intact and without any signs of extravasation. The stent graft was left *in situ*. The sac and graft were decontaminated mechanically, and irrigated thoroughly with hydrogen peroxide 1.5% solution, after which gentamicin collagen sponges were installed in the sac and around the stent graft. Resection of the distal part of transverse colon

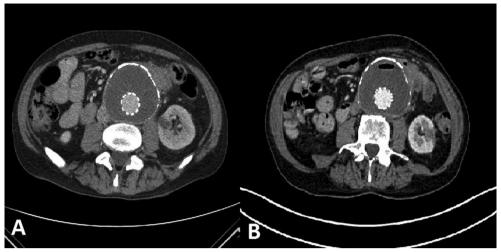


Figure 1. A; CT of Aneurysm sac without air-bubble perigraft in 2013. B; CT of Aneurysm sac with air-bubble perigraft in 2015.

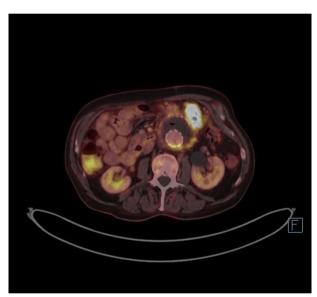


Figure 2. PET-CT scan showing increased focal uptake anterolateral in the aneurysm sac.

was performed and a transverse colostomy was established. The post-operative course was uneventful and histopathological investigations demonstrated xanthogranulomatous inflammation and no malignancy in the resected colon. Bacterial investigation of the purulent material from the sac demonstrated *E. faecium* with an identical resistance pattern to the pre-operative blood cultures. Within 4 post-operative weeks, and in close co-operation with microbiology department, the patient received broad spectrum antibiotic therapy with meropenem, linezolid, vancomycin, and moxifloxacin, and infection parameters normalized completely.

The patient was discharged in good condition with intended lifelong oral doxycycline treatment. The colostomy had been reversed and at the latest follow up (12 months) the patient was without any signs of infection with normal inflammatory parameters and a stable stent graft on CT.

DISCUSSION

AEF is an abnormal communication between the aorta and adjacent intestine and can be primary, or, more common, secondary to aortic surgery.¹

The incidence of AEF after EVAR with an intact aortic wall is unknown but is less than after open AAA repair.²

The clinical manifestation of AEF can be very variable and, depending on the site of the fistula, as diverse as hematemesis, lower gastrointestinal bleeding, sepsis, unexplained fever, or diffuse abdominal pain.³ Onset with endocarditis has, to the best of the authors' knowledge, not been reported previously.

The diversity in clinical presentation of AEF makes the diagnosis challenging, with a possible hazardous diagnostic delay of this often fatal condition.

The interval between EVAR and AEF diagnosis is variable, but is reported to be between 4 and 120 months. ^{4,5} In the present case, the diagnosis was established as late as 168 months (15 years) after EVAR, which is probably among the longest post-operative intervals yet to be reported.

The mechanism underlying the AEF formation to the transverse colon in this otherwise uncomplicated EVAR case is unclear, with no signs of endoleak, migration, or kinking of the graft. Intensive antibiotic treatments can probably explain why the pre-operative bacteriological findings differ from the post-operative findings.

Stent graft infection is an unusual but life threatening condition, with a reported incidence around 1%. ^{6,7} Leaving the stent graft *in situ* instead of a more extensive approach, including graft removal and anatomical or extra-anatomic reconstruction, is debatable, and the conservative approach with preservation of the stent graft, as in the present case, has been reported previously, albeit with disappointing results, with an overall mortality of 45%. ⁸

Extensive surgery was avoided owing to the operative findings of an intact endograft and because of the severe cardiac comorbidity with LVEF of only 20%.

18 M.M.K. Kadhim et al.

CONCLUSION

This case indicates that even uneventful EVAR can generate an AEF in an otherwise successfully excluded and stable aneurysm sac, and that several years of uncomplicated EVAR surveillance is no guarantee against AEF formation. The unforeseen presentation as endocarditis underlines the need for awareness of unusual symptoms and signs that could represent potential late EVAR related complications.

CONFLICT OF INTEREST

None.

FUNDING

None.

REFERENCES

- 1 Mathias J, Mathias E, Jausset F, Oliver A, Sellal C, Laurent V, et al. Aorto-enteric fistulas: a physiopathological approach and computed tomography diagnosis. *Diagn Interv Imaging* 2012;93: 840—51.
- 2 del Moral L, Alonso S, Kiuri S, Caballero D, Heredero A, Nistal M, et al. Aorto-enteric fistula arising as a complication of

- endovascular treatment of abdominal aortic aneurysm. *Ann Vasc Surg* 2009;**23**:255.e13—7.
- 3 Tagowski M, Vieweg H, Wissgott C, Andresen R. Aorto-enteric fistula as a complication of open reconstruction and endovascular repair of abdominal aorta. *Radiol Res Pract* 2014:**2014**:1–6.
- 4 Bergqvist D, Björck M, Nyman R. Secondary aorto-enteric fistula after endovascular aortic interventions: a systematic literature review. J Vasc Interv Radiol 2008;19:163—5.
- 5 Lindblad B, Holst J, Kölbel T, Ivancev K. Malmö Vascular Centre Group. What to do when evidence is lacking—implications on treatment of aortic ulcers, pseudoaneurysms and aorto-enteric fistulae. *Scand J Surg* 2008;97:165—73.
- 6 Davila V, Stone W, Duncan A, Wood E, Jordan W, Zea N, et al. A multicenter experience with the surgical treatment of infected abdominal aortic endografts. *J Vasc Surg* 2015;62:877—83.
- 7 Fatima J, Duncan A, de Grandis E, Oderich G, Kalra M, Gloviczki P, et al. Treatment strategies and outcomes in patients with infected aortic endografts. J Vasc Surg 2013;58:371—9.
- 8 Moulakakis K, Sfyroeras G, Mylonas S, Mantas G, Papapetrou A, Antonopoulos C, et al. Outcome after preservation of infected abdominal aortic endografts. *J Endovasc Surg* 2014;**21**:448—55.