Novel Management of Peri-Aortitis after Endovascular Repair of Abdominal Aortic Aneurysm

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WHAT THIS PAPER ADDS
Peri-aortitis is a rare but significant complication after endovascular aortic aneurysm repair (EVAR). The diagnosis, pathogenesis, and management are poorly understood. In this case the unique diagnosis, management, and follow up algorithm for patients with peri-aortitis after EVAR are presented.

Introduction: The management of peri-aortitis, a rare complication after endovascular aneurysm repair (EVAR) of abdominal aortic aneurysms (AAA), is described in a patient with a solitary kidney.

Report: A 64 year old man who developed peri-aortitis after elective EVAR for a 6.6 cm infrarenal AAA is reported. Peri-aortitis was diagnosed two months after the procedure and was successfully treated with corticosteroids.

Conclusion: There is no clear consensus on the best medical therapy for peri-aortitis secondary to EVAR. Peri-aortitis must be considered in patients with ongoing systemic symptoms of inflammation after EVAR, and early management is crucial to the early resolution of symptoms.

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INTRODUCTION
Peri-aortitis is a rare complication after endovascular aneurysm repair (EVAR) of the aorta. The pathogenesis and treatment are poorly understood. This complication must be differentiated from inflammatory aortic aneurysm, which comprises about 5% of abdominal aortic aneurysms (AAAs) and usually resolves after surgical repair.1 A unique case of peri-aortitis after EVAR and its management in a patient with a solitary kidney is reported.

REPORT
A 64 year old man with a 6.6 cm asymptomatic infrarenal AAA underwent elective EVAR using a bifurcated stent graft (Gore Excluder AAA Endoprosthesis, W.L. Gore & Associates, Flagstaff, AZ, USA). His past medical history included a left sided nephrectomy for trauma, hypertension, and hyperlipidaemia. Pre-operative contrast enhanced computed tomography (CT) revealed no evidence of peri-aortic inflammation (Fig. 1) and the white cell count, renal function, and C reactive protein levels were within normal limits.

A contrast enhanced CT scan performed two months post-operatively demonstrated an increase in the diameter of the aneurysm from 6.6 cm to 7.4 cm. The change in aneurysm diameter was predominantly a result of aortic wall thickening with mild peri-aortic fat stranding along the length of the prosthesis, suggestive of peri-aortitis (Fig. 2).

Inflammatory markers were elevated with a C reactive protein level of 48 mg/L (<5 mg/L) and an erythrocyte sedimentation rate of 80 mm/hour (1–15 mm/hour). He was initially managed conservatively, without starting antibiotics or steroids. However, several months later he presented with abdominal pain and night sweats. A repeat CT scan revealed an interval increase in the adjacent peri-aortitis. A positron emission tomography-computed tomography (PET-CT) scan five months post-intervention revealed intense uptake of 18F-fluorodeoxyglucose throughout the thickened wall of the aneurysm sac, along the length of the prosthesis (Fig. 3A). However, a labelled

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white cell scan demonstrated no abnormal white cell activity around the endograft nor in the fluorodeoxyglucose (FDG) avid thickened aortic wall, which was suggestive of inflammatory peri-aortitis but not of infection of the aortic endograft (Fig. 3B). Subsequently, corticosteroids were initiated using prednisolone (initial dose, 75 mg/day). Follow up renal tract ultrasound performed one, four, and eight months post-operatively revealed no ureteric obstruction. A follow up PET scan was performed after five months of medical therapy and was consistent with excellent partial resolution of the peri-aortitis (Fig. 4). The patient was weaned off steroid therapy gradually.

As part of ongoing surveillance, a CT scan performed 18 months post-operatively revealed a 6 mm annual interval increase in the size of the aneurysm sac. This was found to be secondary to a type two endoleak from the inferior mesenteric artery (IMA). Endoluminal coiling of the IMA was attempted but was abandoned because of anatomical tortuosity. He was subsequently taken to the theatre for laparoscopic stapling of the IMA, and recovered without developing any complications. Laparoscopic assessment of the aorta demonstrated a white, porcelain appearance of the aortic wall (Fig. 5).

DISCUSSION

Peri-aortitis is a rare complication of EVAR that can be challenging to both diagnose and treat. The prompt diagnosis of peri-aortitis after EVAR is vitally important because vascular graft infection must be excluded prior to starting medical treatment. Radiological diagnosis of graft infection may reveal ectopic gas, peri-graft inflammation and fluid, thickening of adjacent bowel, and pseudoaneurysm formation, whereas peri-aortitis generally presents with aortic wall thickening and a low density, mildly enhancing soft tissue mass surrounding the aorta. The extent of fibrosis can be variable and extend to cause ureteric obstruction. In the present case, the diagnosis of peri-aortitis could not be made after CT scan and bloods as they were inconclusive. The diagnosis was confirmed when a labelled white cell scan revealed no abnormal uptake along the graft. Five of the eight reported cases of peri-aortitis after EVAR were diagnosed after initially presenting with ureteric obstruction.

Figure 1. The pre-operative baseline computed tomography scan showed no abnormal vascular wall thickening of the abdominal aortic aneurysm.

Figure 2. The computed tomography scan 2.5 months post endovascular aortic aneurysm repair showed mildly enhancing diffuse wall thickening of the abdominal aortic aneurysm sac, with mild peri-aortic stranding.
Figure 3. (A) The baseline fluorodeoxyglucose (FDG) positron emission tomography-computed tomography (PET-CT) (five months after EVAR) shows intense FDG activity corresponding to the irregular thickened wall of the aortic aneurysm sac. (B) The labelled white cell imaging (five months after endovascular aortic aneurysm repair and several days after the FDG PET-CT) showed no white cell activity at the irregular thickened wall of the aneurysm sac, indicating that the intense inflammatory FDG uptake is of non-infective aetiology. (hexamethylpropyleneamine oxime labeled white blood cell HMPAO-WBC) (Single-photon emission computerized tomography SPECT-CT).

Figure 4. After five months of medical treatment, the fluorodeoxyglucose (FDG) positron emission tomography-computed tomography (PET-CT) showed interval significant improvement of the abnormal intense circumferential FDG uptake corresponding to the irregular thickening wall of the aortic aneurysm sac.
obstruction. Review of the five cases with ureteric obstruction revealed that one patient required a nephrectomy and two patients underwent ureteric stenting. There appears to be no clear consensus regarding the treatment of peri-aortitis secondary to EVAR. Of the eight patients reported previously, five were treated with corticosteroids, one was treated with tamoxifen and two were treated with a combination of corticosteroids and tamoxifen. Two of the papers initiated tamoxifen based on the successful treatment of idiopathic retroperitoneal fibrosis by Clarke et al. and Loffeld et al. However, a recent randomised controlled trial has shown that prednisolone is more effective than tamoxifen in preventing relapses of idiopathic retroperitoneal fibrosis.

Considering this patient had a pre-existing solitary kidney, it was felt that it was important to start early treatment of his condition to avoid irreversible kidney damage. PET-CT and a labelled white cell scan were used to both confirm the diagnosis and exclude evidence of infection prior to starting steroid therapy. Renal tract ultrasound surveillance was used to detect evidence of secondary ureteric obstruction and PET-CT was used for surveillance of residual disease as proposed by Vaglio et al. in the treatment of idiopathic retroperitoneal fibrosis. The most recent PET-CT has shown a near complete response to corticosteroid treatment.

**Conclusion**

Peri-aortitis post-EVAR has been associated with both polyester grafts as well as polytetrafluoroethylene (PTFE), as in this report. This rare complication of EVAR must be considered in patients with ongoing systemic symptoms of inflammation after EVAR. Early management may help early resolution of the symptoms and prevent renal impairment.

**CONFLICT OF INTEREST**

None.

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