Q fever aortic infection causing an aortoduodenal fistula after endovascular aneurysm repair

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ABSTRACT
An aortoduodenal fistula is a rare complication of endovascular aortic aneurysm repair. Q fever infection is known for its vascular tropism, and arterial fistulas have been reported in association with Coxiella burnetii infections. We report the case of a 78-year-old patient who had developed an aortoduodenal fistula secondary to vascular Q fever 5 years after he had been treated with an aortic endograft. Explantation of the endograft, autogenous reconstruction using the neo-aortoiliac system procedure, and duodenal repair were performed as a curative surgical treatment of this serious vascular condition. At the 9-month follow-up examination, the patient showed no signs of recurrent vascular infection and was instructed to complete an 18-month antibiotic regimen. (J Vasc Surg Cases and Innovative Techniques 2020;6:487-9.)

Keywords: Aortoduodenal fistula; Coxiella burnetii; Endovascular treatment; Q fever; Vascular infection

The development of an aortoenteric fistula (AEF) after aortic endovascular repair (EVAR) is a challenging condition and has been described secondary to endoleaks, graft migration, and intra-abdominal inflammatory processes. Coxiella burnetii, causing Q fever infection, is known for its affinity for cardiovascular structures and has resulted in infection of aneurysms and vascular grafts. We report the case of an AEF caused by an infected aneurysm with Q fever arising 5 years after EVAR. Definitive surgical treatment included graft explantation and in situ autogenous reconstruction. The patient agreed to publish this report.

CASE REPORT
A 78-year-old man who had undergone uneventful EVAR of an 8-cm fusiform aneurysm (Endurant; Medtronic, Dublin, Ireland) had presented 5 years later at his annual follow-up examination with recent ultrasound imaging findings suggestive of a proximal type Ia endoleak. What had been reported as a type I endoleak on the ultrasound scan was more precisely defined as a 5-cm heterogeneous collection coming into intimate contact with the irregular wall of the third duodenum (Fig 1), which had initially been described as a type I endoleak. Gastroscopy revealed a 1-cm distal duodenal ulcer without visualization of the endograft (Fig 2). A positron emission tomography scan confirmed an intense hypermetabolic area surrounding the infrarenal aorta and endograft, compatible with graft infection (Fig 3). These findings were consistent with an aortoduodenal fistula.

He did not complain of any abdominal or lumbar pain nor did he have any gastrointestinal symptoms. His vital signs and physical examination findings were unremarkable. Laboratory studies revealed a hemoglobin level of 11.9 g/dL, normal white blood cell count, and creatinine level of 0.83 mg/dL. Inflammatory markers were slightly elevated (erythrocyte sedimentation rate, 33 mm/h; C-reactive protein, 13 mg/L).

Computed tomography angiography showed a 7-mm increase of the aneurysm sac and a disruption of the aortic wall at the level of the infrarenal neck. What had been reported as a finding suspicious for a type I endoleak on the ultrasound scan was more precisely defined as a 5-cm heterogeneous collection coming into intimate contact with the irregular wall of the third duodenum (Fig 1), which had initially been described as a type I endoleak. Gastroscopy revealed a 1-cm distal duodenal ulcer without visualization of the endograft (Fig 2). A positron emission tomography scan confirmed an intense hypermetabolic area surrounding the infrarenal aorta and endograft, compatible with graft infection (Fig 3). These findings were consistent with an aortoduodenal fistula.

A prompt preoperative workup was performed, and the patient was deemed operable for endograft explantation, neo-aortoiliac system (NAIS) reconstruction, and duodenal repair. With the patient under general anesthesia, a bifurcated graft was created using bilateral suprapopliteal femoral veins. Midline laparotomy was then performed to expose the supraceliac aorta to obtain proximal control. The infrarenal aorta was dissected, which revealed the third portion of the duodenum plastered to the aneurysm sac. The supraceliac aorta was clamped. The opening of the aneurysm sac revealed an abscess into the arterial wall and a small, but direct, communication between the third portion of the duodenum and the midportion of the aneurysm. Cultures and biopsies of the infected aneurysm were taken. The entire endograft was easily explanted, and no metallic bars were seen coming through the aortic wall to explain the fistula. The NAIS procedure was performed, and
the small duodenal defect was primarily repaired and secured using an interposed omental flap.

His postoperative course was favorable, and the patient was discharged home on day 10. The patient tested positive for Q fever with phase I and II IgG antibody titers of 1:2048 by immunofluorescence assay (chronic Q fever considered present if phase I IgG level >1:800). The final pathologic examination of the biopsy specimens taken during the procedure revealed necrotizing granulomas, proving the presence of aortic infection. The perioperative cultures also revealed gastrointestinal flora; therefore, intravenous broad-spectrum antibiotics were continued for 6 weeks. For treatment of the vascular Q fever, the patient will receive a doxycycline and hydroxychloroquine regimen for a total of 18 months. At the 9-month follow-up examination, the patient was doing well, and a computed tomography angiogram showed patent vascular reconstruction without signs of residual infection.

**DISCUSSION**

Secondary AEF after open aneurysm repair is a rare, but well-described, late surgical complication. It has been reported in $\leq$1.6% of cases. Meticulous closure of the aneurysm sac and retroperitoneal space is paramount to avoid the development of such a morbid condition. Interposition of viable tissue should be considered. The less invasive use of endovascular technology avoids dissection of the duodenum and suture line exposure and isolates the synthetic material within the aortic lumen, avoiding the classic factors associated with the development of a fistula. However, after EVAR, AEFs have been reported. The causes have included graft migration and kinking, sac expansion with or without an endoleak, an inflammatory aneurysm, inflammatory bowel disease, and metallic stents protruding through the aortic wall with adjacent organ injury. The MAEFISTO study (multicenter study on aortoenteric fistulization after stent grafting of the abdominal aorta) evaluated 3932 endograft implantations from 1997 to 2013 and reported an incidence of AEF development after EVAR of 0.46% for atherosclerotic aneurysmal disease and 3.9% when performed for postoperative pseudoaneurysm. The findings from the present case have shown that an aneurysm sac infection can also be a potential cause of AEF after EVAR. In our patient, EVAR had been performed 5 years before the clinical presentation of
infection and AEF, and the findings from regular postoperative imaging studies and clinical follow-up examinations had been unremarkable. Also, at graft explantation, no metallic barbs had protruded through the aortic wall, and the orifice of the fistula was found to be in the middle segment of the excluded aneurysm, with the endograft deeply buried within the thrombosed aneurysm sac. Because Q fever has been increasingly diagnosed in our vascular center, we now routinely test all patients with a vascular infection for Q fever. Also, because the present patient lives close to a sheep farm, we believe he had developed chronic Q fever infection of the aneurysm sac that led to the development of the AEF.

Q fever is a zoonosis caused by *C. burnetii*, a pathogen with a vast animal reservoir, including as sheep, goats, and cattle. Q fever can present as either an acute or chronic infection, with the latter a rare condition developing in 1% to 5% of patients. Chronic Q fever infection can develop months to years after the index exposure to the pathogen, which is known for its tropism for vascular structures. Endocarditis has been the most frequent vascular manifestation of chronic Q fever, followed by aneurysm and vascular graft infection. Infected aneurysms can present with necrotizing granulomas, which will infiltrate and weaken the vessel wall, leading to accelerated aneurysm growth and an enhanced risk of rupture. In addition, 15% of patients with chronic Q fever presenting with vascular involvement will have an arterial fistula, and the mortality rate has been high, even if treated surgically. A few cases of Q fever–related AECs have been previously reported.

Just as with other vascular infections, the surgical treatment of a vascular Q fever requires control of the source of the infection, removal of all synthetic material, and restoration of vascular continuity. Although graft-preserving techniques have been described in the treatment of Q fever infection, we favored complete graft explantation considering the intense uptake found on the positron emission tomography scan. The NAIS procedure was used for revascularization because it is known for its durability and low rate of reinfection.

**CONCLUSIONS**

Chronic Q fever is rare but can result in major vascular complications, including AEF formation after EVAR. Prompt diagnosis is of utmost importance, and Q fever serology should be a part of the routine vascular infection workup. Complete explantation of the graft and autogenous reconstruction combined with a long-term antibiotic regimen can lead to favorable outcomes.

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