David procedure during a reoperation for ongoing chronic Q fever infection of an ascending aortic prosthesis

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Abstract
Chronic Q fever infections, caused by Coxiella burnetii, are associated with cardiovascular complications, mainly endocarditis and vascular (graft) infections. We report a case of a patient with a C. burnetii infected thoracic aorta graft treated initially in a conservative way. However, surgical excision of the infected graft was eventually necessary. This case report highlights the challenges regarding the treatment of patients with chronic vascular C. burnetii infections. In the absence of practical guidelines, treatment is tailored to the individual patient. Furthermore, we want to emphasize the need to include chronic Q fever in the differential diagnosis in patients with culture negative aortitis, especially in the regions with Q fever epidemics in the recent past.

Keywords: Q fever • Vascular graft • Chronic infection

CASE REPORT
A 55-year old woman was seen in a peripheral hospital with complaints of weakness, fatigue, dyspnoea and a continuous chest pain for 3 weeks. Her medical history revealed a supracoronary tube graft repair of the ascending thoracic aorta for a type A aortic dissection 3 years earlier. A physical examination revealed no abnormalities. The C-reactive protein (CRP) was 18 mg/l, leukocyte count was $6.3 \times 10^9$/l and erythrocyte sedimentation rate (ESR) was 15 mm/h. Liver and kidney functions were in normal ranges, apart from minor elevations in gamma-GT and alkaline phosphatase. Computerized tomography (CT) (Fig. 1A) and fluorodeoxyglucose (FDG)-positron emission tomography (PET)/CT (Fig. 1B) of the thorax showed a fluid collection around the prosthesis and an increased FDG uptake of the aortic wall and stent graft, indicating suspected aortitis and eventual prosthetic graft infection. The transthoracic echocardiogram showed normal functioning valves without vegetations. Microbiological screening, including repeated blood cultures, was negative. However, in the work-up for culture negative aortitis, Coxiella burnetii (C. burnetii) serology was positive with elevated IgG phase I and II titres (both 1:4096), indicative of a chronic Q fever infection. A C. burnetii specific polymerase chain reaction (PCR) test was negative in plasma. On the basis of the relatively good clinical presentation of the patient, it was initially decided to treat the patient conservatively with antibiotics (doxycycline and hydroxychloroquine). Microbiology parameters, including C. burnetii PCR and serology were checked every three months.

Despite adequate antibiotic serum levels for almost a year, no clinical improvement was documented: the patient still complained of weakness, fatigue and a continuous chest pain. Subsequent CT scans demonstrated augmentation of the fluid mass surrounding the ascending aorta tube graft. IgG levels remained high and after a year a C. burnetii PCR was positive in the fluid material obtained by a diagnostic puncture as well as in a peripheral blood sample.

Treatment options were again discussed and determined in a multidisciplinary setting. On the basis of the non-responsiveness to conservative treatment and even progression on a CT-scan, we decided to treat the patient surgically as well. The operation was performed using cardiopulmonary bypass through the femoral vein and artery. Immediately after a median sternotomy, pus surrounding the prosthesis was visible. After removal of the pus, an aortic clamp was placed and the heart was preserved in crystalloid solution. The infected graft was removed and because of a wide aortic root (40 mm) and a normal functioning aortic valve, a David procedure was performed, using a 28-mm Gelweave graft (Vascutek, UK) soaked in rifampicin. The aortic clamp time was 146 min and the cardiopulmonary bypass time was 231 min. Normothermia was accomplished during the whole procedure. Culture of the infected prosthesis and pus remained negative, but C. burnetii DNA was detected by PCR in this material. Moxifloxacin was added to the antibiotic treatment until major side effects urged us to minimize the treatment to moxifloxacin only, 3 months after surgery. Except for drainage of pleural effusion, the patient had a good recovery and was...
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Figure 1: (A and B) Preoperative CT scan showing fluid collection around the ascending aorta after a tube graft repair (arrow) with expansion to the retrosternal space (arrowhead) (A). Peri-aortitis was suggested clearly by a PET–CT showing elevated FDG uptake of the aortic wall and stand graft of the ascending aorta (arrow) (B).

DISCUSSION

Q fever is a zoonosis, caused by the Gram-negative coccobacillus C. burnetii. There is a large animal reservoir, with goats, sheep and cattle being the most common source of human infections [1]. From 2007 till 2009, the Netherlands suffered a major Q fever outbreak, with more than 4000 reported symptomatic cases. As the majority of patients show mild or asymptomatic acute infections, the actual incidence is probably much higher [2]. After primary infection, an estimated 1–5% of the patients progress to chronic Q fever, which can become manifest years after the initial infection. Endocarditis and infections of aneurysms or vascular prostheses are the most common manifestations [3]. Pre-existent cardiac valvular disease, an aortic aneurysm, vascular grafts and/or an immunocompromised state are reported risk factors for the development of chronic Q fever [3]. The diagnosis of chronic Q fever relies mainly on the serology and detection of DNA in blood or tissue with PCR. Chronic Q fever is proved when C. burnetii DNA is detected by PCR in blood or tissue, although the sensitivity of these techniques in peripheral blood is low [4]. Persisting high levels of antibodies to phase I and phase II antigens are also considered indicative of chronic Q fever [4]. The clinical features of patients with chronic Q fever are non-specific findings such as fatigue, fever, abdominal or chest pain, weight loss, night sweats and hepato-splenomegaly [3]. In the presented patient, only fatigue and chest pain were mentioned. Infection parameters, including CRP, leucocyte count and ESR results, were only mildly elevated. This, combined with the fact that most patients with a manifest chronic Q fever infection are unaware of a primary infection with C. burnetii, contributes to a significant diagnostic delay. If left untreated, chronic Q fever leads to considerable morbidity and even mortality in up to 60% of cases [3]. Long-term antibiotic treatment, preferably consisting of hydroxychloroquine and doxycycline, is required [3]. However, no comparative trials regarding antibiotic and/or surgical treatment for chronic Q fever are available. In our patient, treatment with both mentioned antibiotics resulted in no clinical improvement. It is plausible that this failure was based on acquired resistance during long-lasting antibiotic treatment or as a result of the fact that the infection was situated at a polyester prosthesis in an anatomical position with a low antibiotic penetration possibility. Several surgical problems are encountered in patients with infected grafts. Owing to tissue damage, bleeding is of major concern. Therefore, cardiopulmonary bypass is needed during the procedure, which can be achieved by cannulation of the patient through the femoral artery and vein before opening the sternum. Tissue around the infected prostheses can be fragile, making it more difficult to fix a new graft [5].

In conclusion, we recommend the inclusion of chronic Q fever in the differential diagnosis of patients with culture negative aortitis, especially in regions with Q fever epidemics in the recent past. Owing to the deficiency of clinical and microbiological data, the criteria for surgical (re-) intervention are not straightforward and should be tailored to the individual patient.

Conflict of interest: none declared.

REFERENCES


discharged after 3 weeks. A control CT showed a significant decrease of the peri-aortic and retrosternal fluid collection. Six months postoperatively, the patient remains alive and well.