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*Case Report*

# Vertebral Osteomyelitis or Infected Abdominal Aortic Endograft? A Rare Case of Q Fever

Q2Q1

Q3Q7 *V. Waelbers,[1] S. Desmet,[2] P. De Munter,[3] J. Van Loon,[4] and I. Fourneau,[1] Leuven, Belgium*

*Coxiella burnetii* is the etiological agent of Q fever, a zoonosis. Vascular infections are associated with significant morbidity and mortality. Osteoarticular Q fever infections are rare. We describe a case of vertebral osteomyelitis with associated infection of an abdominal aortic endograft, caused by *C. burnetii*. Most probably, an initial pyogenic vertebral osteomyelitis extended locally to the endograft. Treatment consisted of antibiotic therapy and surgical resection of the infected aortic endograft and in situ reconstruction with autogenous superficial femoral vein grafts.

## INTRODUCTION

*Coxiella burnetii* is an intracellular, gram-negative bacterium which causes Q fever, a zoonotic disease. It is prevalent globally, especially in rural areas. The primary source of infection is farm animals, and transmission occurs mainly through inhalation of contaminated aerosols.[1,2]

Clinical presentation of the disease is polymorphic, and symptoms are nonspecific. Acute Q fever is asymptomatic in 60% of cases but can present with isolated fever, atypical pneumonia, and hepatitis. 1–5% of all patients will develop chronic Q fever, manifesting as endocarditis (60–80%), vascular infections (9%), and infectious complications during pregnancy (5%).[2,3] Osteoarticular infections are rare.[3,4]

We describe a case of vertebral osteomyelitis with associated infection of an abdominal aortic endograft caused by *C. burnetii*.

## CASE REPORT

A 69-year-old woman was admitted to the hospital in February 2018 with an acute exacerbation of lower back pain and fever. Medical history included an elective endovascular aneurysm repair in August 2000 for an infrarenal abdominal aortic aneurysm. She presented with spinal tenderness without neurological deficits. The laboratory findings showed an elevated C-reactive protein of 129.9 mg/L (normal <5 mg/L) and leukocytosis of $4.2 \times 10^3/\mu L$ (normal $3.5–11 \times 10^3/\mu L$). Computed tomography (CT) with contrast demonstrated a presacral hypodense collection, reaching up against the aortic bifurcation, suspicious for abscess (Fig. 1). There was also osteolytic damage to vertebrae L4–L5. She was empirically treated with intravenous amoxicillin-clavulanic acid, which was changed after 2 days to piperacillin-tazobactam because of persistent high fever. She was subsequently transferred to the vascular unit of our tertiary referral hospital. Blood cultures were found to be negative and the patient was afebrile, so antibiotics were stopped.

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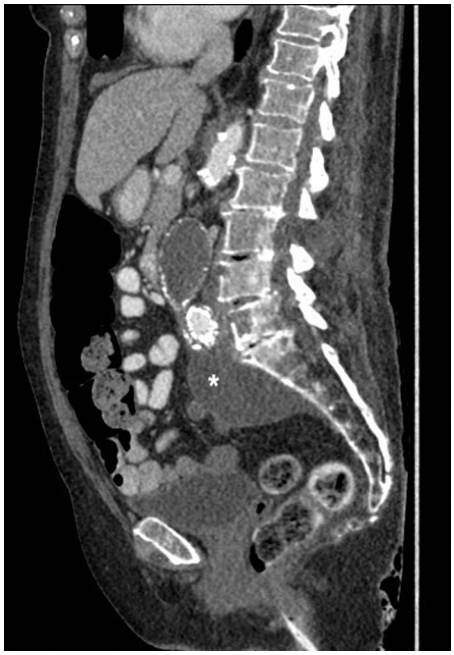


**Fig. 1.** Computed tomography showing a presacral hypodense abscess collection (*).

Magnetic resonance imaging showed advanced vertebral osteomyelitis of L3−L5 with destruction of the anterior part of L4 and L5. There were abscess collections in these vertebral bodies with an epidural expansion from the intervertebral spaces. Furthermore, there was an additional voluminous central necrotic mass prevertebral from L5 to S3 with contrast-staining wall and septa in close relation to the aortoiliac endoprosthesis (Fig. 2). Positron emission tomography (PET)/CT confirmed the vertebral osteomyelitis with epidural expansion and an associated surinfection of the aortic endoprosthesis limited to the level of the bifurcation and proximal right iliac prosthesis with a presacral abscess collection (Fig. 3).

A CT-guided puncture biopsy of the vertebra L4 was performed. The culture turned negative. A panbacterial polymerase chain reaction (PCR) (16S rRNA PCR with amplicon sequencing) was positive for *Staphylococcus* species without further typing at a species level, presumably a contaminant. In March 2018, an open biopsy through a left interlaminar access, L3−L4, was performed along with drainage of purulent fluid. The cultures remained negative, but the panbacterial PCR was positive for *C. burnetii*. Serological analysis for Q fever revealed significant titers (phase I IgG 1/4096 and phase II IgG 1/16384, phase I IgM >1/2048 and, phase II IgM 1/1024). Inquiring with the patient taught that she indeed has contact with sheep living behind her house. Therapy with doxycycline (200 mg/day) and hydroxychloroquine (600 mg/day) was initiated. A transesophageal cardiac ultrasound was carried out twice, demonstrating no cardiac valve lesions.

The aortic endoprosthesis was considered as a maintaining factor for the infection. Therefore, the aortic endoprosthesis was removed in April 2018, and in situ

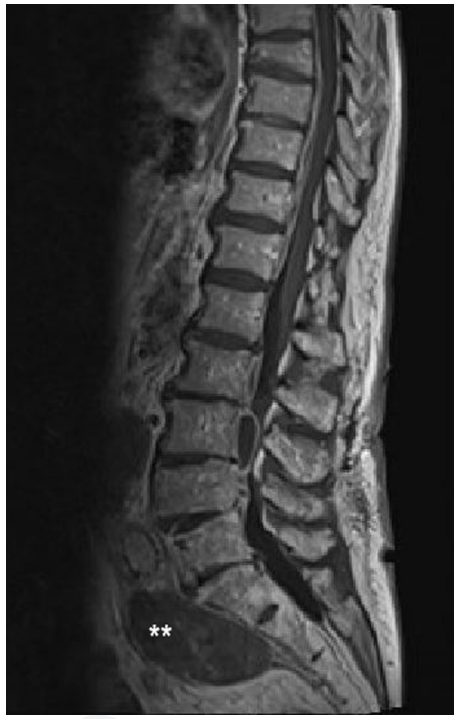


**Fig. 2.** Magnetic resonance imaging: contrast-enhanced T1 (T1c) image, demonstrating spondylodiscitis of L3−L5 with epidural and prevertebral abscess collections (**).

reconstruction was performed by an autogenous venous aorto-bi-iliac graft, using the superficial femoral veins. Bacteriological cultures from the aortic wall and endoprosthesis remained negative. The patient's recovery was uneventful except for a hypo-osmolar hyponatremia which was corrected, and she was discharged to home on day 31.

A CT after 3 months showed postoperative changes around the venous aorto-bi-iliac graft without any signs of infection. Five months after surgery, the patient remains well with decreasing Q fever titers (phase I IgG 1/1024, phase II IgG >1/2048, phase I IgM >1/512, phase II IgM 1/128). Serum levels of leukocytes and C-reactive protein were normalized. Antibiotics will be continued for at least 18 months under serologic monitoring.

## DISCUSSION

Q fever is a zoonosis caused by *C. burnetii*, an obligate intracellular gram-negative bacterium. It is prevalent in almost every country except New Zealand. Most infections are the result of inhalation of air contaminated by the excreta of infected animals. Cattle, sheep, and goats are primary reservoirs, but other mammals, birds, and anthropods have also been implicated.[2,3]

Primary infection is asymptomatic in 60% of cases but can lead to acute Q fever with a pleomorphic presentation. It can range from myalgia,

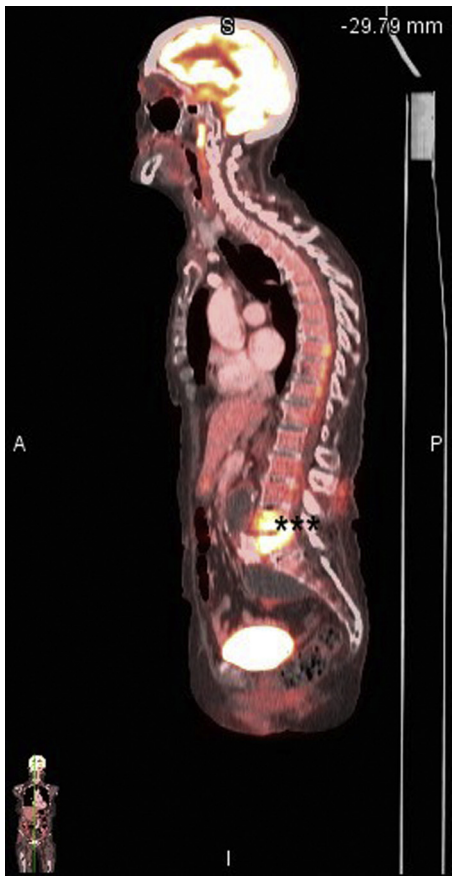


**Fig. 3.** Positron emission tomography/CT fusion shows spondylodiscitis and associated surinfection of the aortic endoprosthesis (***).

headache, or fever to atypical pneumonia, hepatitis, meningitis, or myocarditis. Acute infection can progress to chronic Q fever with endocarditis as its most frequent clinical presentation (60–80%). Vascular infections are the second most common form (9%).[2,3] *C. burnetti* has an affinity for cardiovascular tissues and vascular grafts and is able to persist in monocytes and macrophages that are present in aortic thrombus and damaged cardiac valves.[5] Other presentations, such as musculoskeletal infections and chronic hepatitis are rare.[3,4]

The present report describes a case with vertebral osteomyelitis and aortic endograft Q fever infection. The coexistence of aortic graft infection and osteomyelitis has been reported in several cases, all after open surgical repair of abdominal aortic aneurysms. These were not considered as pure osteoarticular infections because they were caused by extension from the infected graft that directly overlay the vertebra that was involved.[6,7] Piquet et al. successfully treated a patient with contiguous vertebral osteomyelitis caused by extension from the infected pseudoaneurysm of an aortic graft.[8] Such local extension has also been reported by Stokes et al., who described a case of *C. burnetii* infection of an aortic graft with concomitant vertebral osteomyelitis L1–L3.[9]

Osteoarticular Q fever infections are rare.[3,4,11] Landais et al. reported two cases of Q fever spondylodiscitis complicated by paravertebral abscess.[11] To our knowledge, this is the first reported case of Q fever with an infected aortic endograft and vertebral osteomyelitis. A case in which the aortic endoprosthesis was complicated by an ilioenteral fistula and pyogenic vertebral osteomyelitis with *Escherichia coli* have been described by de Koning et al.[12] Kloppenburg et al. described a *C. burnetii* infection of the wall of an abdominal aortic aneurysm in a patient with a previous endovascular aortic aneurysm repair. They performed a resection of the aneurysm wall and left the noninfected endograft in place as a bailout procedure.[13]

It remains unclear where the primary infection focus was located in our patient. The vertebral osteomyelitis is possibly due to direct extension from the adjacent infected periaortic tissue. In this way, the primary infection of the endograft caused erosion of the vertebral bodies. But on PET/CT, there was a very strong uptake of $^{18}$F-fluorodeoxyglucose (FDG) at the anterior cortex of L4–5 and the prevertebral soft tissues with only a mild FDG uptake locally at the bifurcation level of the aortic endoprosthesis. Focal infection of an aortic graft is also very rare. Therefore, most probably, there was an initial pyogenic vertebral osteomyelitis which locally extended to the endograft.

Chronic *C. burnetii* infections of aneurysms and vascular grafts are associated with significant mortality. Antibiotic treatment without surgery cannot be considered a definite treatment in most patients because of the high risk of persistent infection. Wegdam-Blans et al. reported a mortality rate of 70% in patients managed without surgery.[14] Still, surgical repair is also associated with considerable morbidity and mortality. In the same review, the major complication and mortality rate was 21% and 18%, respectively, after surgery in 40 chronic Q fever patients.[14] One study reported significant improvement of recovery, when surgery was performed shortly after the diagnosis of chronic Q fever.[15]

Surgical excision of the infected graft or aneurysm is mandatory. A graft replacement in anatomic or extra-anatomic configuration can be performed.[7,8] In our case, we used the autogenous superficial femoral veins as they are associated with a low risk of reinfection. Alternatively, reconstruction using an autologous great saphenous spiral

vein graft has been successfully performed.[16] It is unknown which surgical technique is preferable for vascular reconstruction, and the choice should be based on the experience of the surgeon.

No specific guidelines are available for the antibiotic treatment of vascular Q fever patients. The current antibiotic regimen of choice is doxycycline and hydroxychloroquine for at least 18 months.[17,18] Hydroxychloroquine increases lysosomal pH to selectively enhance the bactericidal activity of doxycycline.[18]

## CONCLUSION

*C. burnetii* vascular infections are associated with significant morbidity and mortality. We described a rare case of Q fever vertebral osteomyelitis with associated contiguous infection of an abdominal aortic endograft. It was treated successfully with doxycycline, hydroxychloroquine, and replacement with autogenous superficial femoral vein grafts. We emphasize the need for resection of the infected vascular graft together with administration of long-term antibiotic therapy as treatment for chronic vascular Q fever.

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