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Case report

A rare case of aortic endograft infection by *Francisella tularensis*: A case report

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ABSTRACT

Introduction and importance: endovascular repair is an alternative to open repair for abdominal aortic aneurysms (AAA), which lowers morbidity and mortality but may presents infectious complications. Endograft infection is a rare but serious life-threatening condition with a mortality rate up to 50 %. We reported a case of aortic endograft infection by *Francisella tularensis*, rare and highly virulent gram-negative coccobacillus known for use in bioterrorism.

Case presentation: A 79-year-old man presented with asthenia, weight loss, night sweats and one episode of fever. In 2007, he underwent aorto-bi-iliac endograft repair for AAA without any complication. The diagnostic workup showed some signs of inflammation, but negative blood cultures and no sign of infection on CT scan. The combination of positron emission tomography (PET) and white blood cell (WBC) scintigraphy led to the diagnosis of aortic endograft infection. The management was antimicrobial therapy and surgery. Perioperative analysis shows the presence of *Francisella Tularensis*.

Discussion and conclusions: Aortic endograft infection is a serious complication with a high mortality rate. Its diagnosis may be difficult, but the combination of WBC scintigraphy and PET scan may improve identification of the infection, even if blood cultures and CT scan are negative. The gold standard treatment is removal of the endograft, debridement, and in situ reconstruction along with antibacterial therapy.

1. Introduction

Endovascular aneurysm repair (EVAR) is an alternative to open repair (OR) for patients with abdominal aortic aneurysm (AAA) [1]. Postoperative complications can be frequent (16 %–30 %), may require reintervention in up to 19 % of cases, and can be life-threatening [2]. Widespread EVAR use has increased infectious complications [3]. Aortic endograft infection (AEGI) occurs in <1 % of EVAR cases with high mortality rates (25 %–50 %) [4]. The diagnosis of AEGI may be challenging because of nonspecific symptomatology, possibly negative computed tomography (CT) scan and negative cultures in up to one-third of cases [3]. The gold standard management is complete removal of the endograft, debridement and reconstruction. However, conservative treatment, such as percutaneous, or surgical drainage or life-long antibiotic therapy, can be acceptable for patients with high surgical risk [3].

We reported a case of AEGI by *Francisella tularensis*. This work was reported in line with the SCARE criteria [5].

To our best knowledge, this is the first documented case, in patient referred to our University Hospital, of AEGI with *F. tularensis* which is a highly virulent gram-negative coccobacillus that causes a rare bacterial zoonotic disease and may be transmitted to humans by multiple ways (Contaminated water or food ingestion; arthropod or animals bites; direct contact with infected animals, animal tissues, or fluids; or inhalation of aerosols) [6,7]. The incidence of tularemia varies widely. In Europe, only 800 cases was reported annually [8]. Tularemia manifests as an ulceroglandular, glandular, oculoglandular, oropharyngeal, pneumonic, or typhoidal disease [6]. Reported cases of pericarditis, meningitis, and endocarditis had been rare, with only two cases in Europe and two in the United States, probably because of difficulties in

Abbreviations: AAA, abdominal aortic aneurysm; CAG, cryopreserved allograft; AEGI, aortic endograft infection; EAR, extraanatomic reconstruction; EVAR, endovascular aneurysm repair; ISR, in situ reconstruction; OR, open repair; CT, computed tomography scan; CTA, computed tomography angiogram; PET, positron emission tomography; PDF, prosthetic–duodenal fistula; PTFE, polytetrafluoroethylene; WBC, white blood cell.

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obtaining positive blood cultures [9].

2. case presentation

A 79-year-old man presented with asthenia, weight loss for 1 month, night sweats and only one fever episode. There were systemic symptoms. He had no recent travel. He lived in the countryside. He had no animals or history of hunting, fishing, or contact with animals or their feces. He had contact with a dead fox 1 year ago.

From his medical history, we noted arterial hypertension, sleep apnea syndrome, coronary artery disease treated by coronary stents in 2007 and coronary bypass in 1999. In 2007, he underwent EVAR with aorto-bi-iliac endograft (Gore Excluder) for 56 mm AAA; there was no complication. The patient remained asymptomatic during the follow-up period with progressive decrease in aortic aneurysm size to 39 mm in 2010.

His physical examination was normal. Blood tests showed minor signs of inflammation. The blood and urine cultures and serology tests were negative. Gastroscopy, colonoscopy echocardiography, chest radiograph and thoracoabdominal CT scan were normal.

99mTc-Hexamethylpropyleneamine oxime-labeled WBC scintigraphy (Fig. 1) showed accumulation of labeled WBCs at three spots. One of them, localized at the aortoiliac bifurcation, was suspected to be a zone of quiet infection. Positron emission tomography (PET) (Fig. 2) favored inflammation or AEGI. Based on those combined findings we concluded to aortic endograft infection and decided to proceed with surgery.

A midline laparotomy was performed. We exposed the abdominal aorta and iliac arteries. The tissues were inflamed with numerous lymph nodes. After heparin administration, the aorta was clamped below the renal arteries, and the iliac arteries were clamped distally. Upon aneurysmal sac opening, we found around 10 mL of cloudy fluid surrounding the endograft. The aortic sac was freed from the surrounding structures and removed. The two distal endograft limbs were removed from the common iliac arteries and clamped, and the aortic clamp was moved above the renal arteries to explant the endograft. Once the endograft was extracted, we clamped the aorta below the renal arteries (clamping time < one minute). The common iliac arteries and aorta were cleaned and

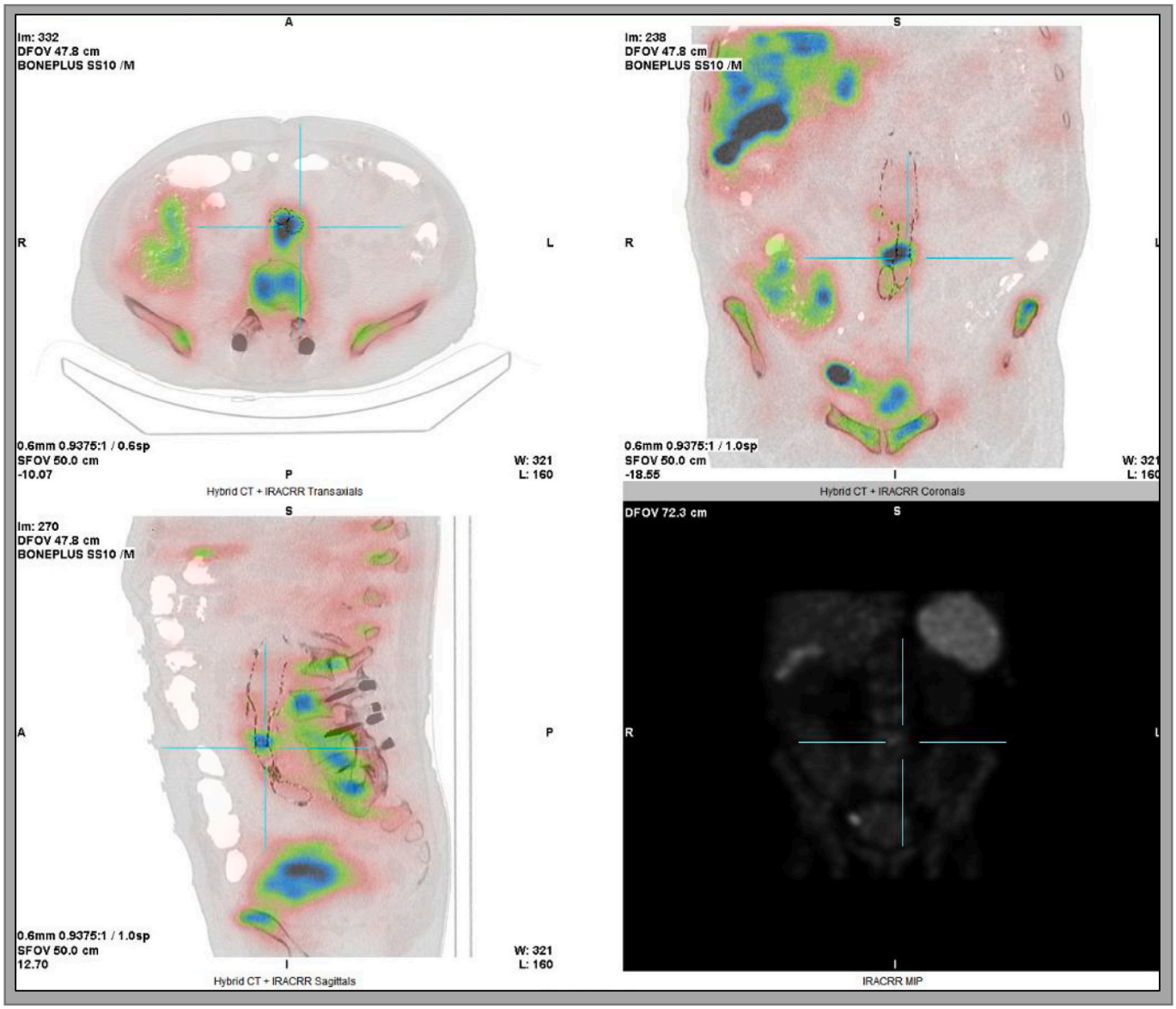

Fig. 1. 99mTc-HMPAO-labeled WBC scintigraphy.
There is accumulation of labeled WBCs at the aortoiliac bifurcation.
WBC, white blood cell.

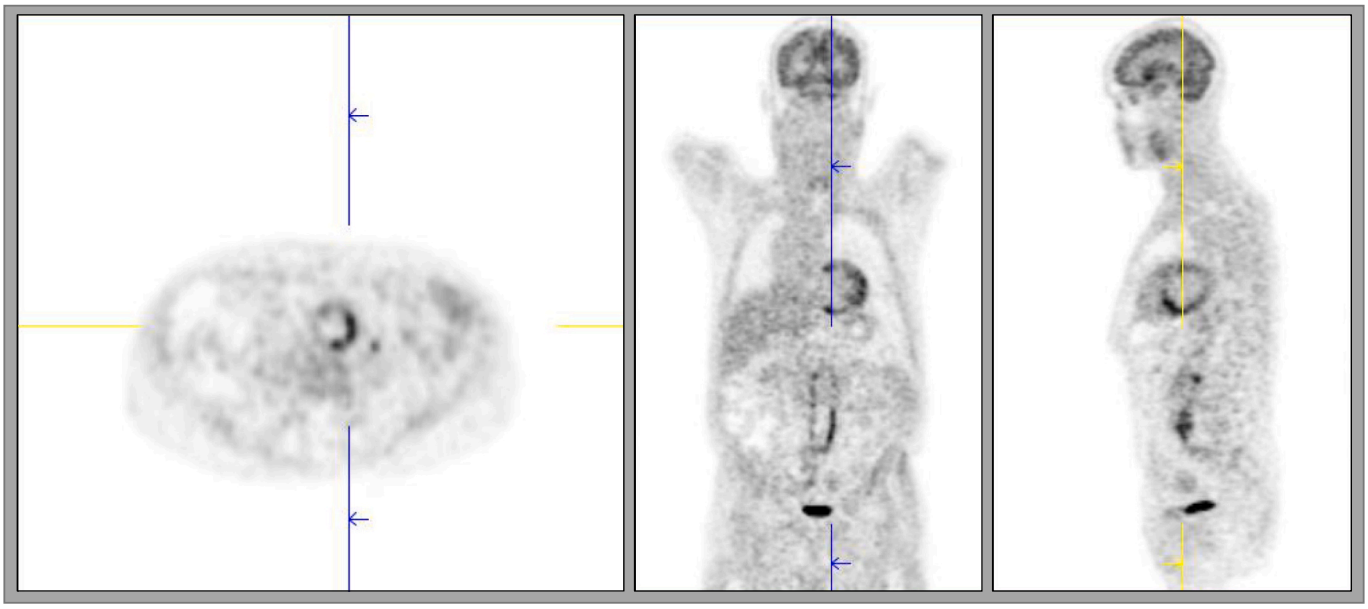

Fig. 2. Positron emission tomography.
The findings favor infectious and/or inflammatory processes in the aortic graft, in relation to the L4–L5 junction.

silver-coated polyesters aortic graft was anastomosed end-to-end to the infrarenal aorta and to the aorta above its bifurcation. Empiric tazobactam and vancomycin were started.

Cultures of the periaortic and aortic tissues showed *F. tularensis*. We shifted the antibiotics to ciprofloxacin and intravenous gentamicin for 2 weeks, followed by ciprofloxacin alone for 10 weeks [10,11]. The inflammatory markers decreased progressively.

On six months follow-up, the patient was doing well without signs of new or recurrent infection.

3. Discussion

F. tularensis is a highly infectious gram-negative coccobacillus. The most recent epidemiological report on tularemia in 2019 showed 1463 cases, with a notification rate of 0.3/100,000 [12]. Only four cases of endocarditis, and one prosthetic valve endocarditis were reported. To our best knowledge, we report the first documented case of EVAR infection with *F. tularensis*.

EVAR has specific complications, such endoleak, iliac branch occlusion and spinal cord ischemia. The high postprocedural complications rate need lifelong imaging surveillance [4].

The risk factors for AEGI are emergency setting of the procedure, the need for surgical revision, use of the radiology suite instead of the operating room, the presence of fever before the procedure, and too short prophylactic antibiotic therapy [3]. AEGI had been associated with endoleaks, percutaneous coiling, and placement of sealant or glue in the aneurysmal sac [3,13]. The mean interval between endograft placement and infection symptoms is 28.3 months [3]. In our case, this interval is much longer (15 years).

Patients usually present fever (71.4 %), back pain (57.1 %), and sometimes weight loss associated with inflammation and leukocytosis in approximately two-thirds of cases [4,14,15].

The most commonly isolated microorganisms in blood cultures are gram-positive cocci (66 %). Only 26.7 % of blood cultures could be positive [3]. CT scan could show mesenteric fat stranding adjacent to the stent graft, perigraft fluid collections, abnormal enhancement, air bubbles and/or adjacent structures erosion [14,16]. Technetium-labeled WBC scanning is useful for aortic graft infection diagnosis, particularly in the low-grade stages [17]. PET can be useful in confirming the diagnosis in highly suspicious clinical situations with negative blood tests and inconclusive CTA [3]. Our patient presents high suspicion of endograft infection with negative blood cultures. PET and labeled WBC scintigraphy were compatible with infection, which was later confirmed by perioperative tissues culture.

The patient's underlying condition, virulence of the infectious organism, the presence of prosthetic–duodenal fistula (PDF) and the AEGI treatment modalities influenced the patient's prognosis. In case of surgical treatment, suprarenal aortic clamping and endograft fixation increased the mortality rate [3]. The different surgical treatment modalities include in situ reconstruction (ISR) or extra-anatomic reconstruction (EAR) [18]. During the 1980s the most common treatment was graft excision with EAR, but this technique has low patency, a high amputation rate and a significant risk of rupture along the aortic suture line [12,19]. To minimize these problems, ISR using different conduits [i.e., autogenous veins, cryopreserved allografts (CAG), synthetic grafts made from polyester/polytetrafluoroethylene (PTFE), and either rifampicin-bonded or silver-coated prosthesis] was introduced [19]. ISR showed lower complication and mortality rates than after EAR [20].

The choice of graft type depends on local protocols and the patient's underlying condition [20]. Although no ideal graft material exists, biological material conduit is preferred because its lowest reinfection rate (<10 %) and higher survival rates [13,15]. An autologous vein might be preferred for young patients [20]. Some teams prefer a cryopreserved arterial homograft ISR because of its capacity to mimic the structural and hemodynamic characteristics of autologous vessels while avoiding the harvesting [3].

Silver/rifampicin polyesters and cryopreserved allograft may be more suitable if PDF is identified. For a low virulence infection, rifampicin-soaked or silver grafts were reported to be effective. For large perigraft abscesses and methicillin-resistant *Staphylococcus aureus* infections, EAR should be considered [20].

Standard expanded PTFE and polyester grafts had significantly worse results for graft occlusion and amputation [10,20]. The reinfection rates did not significantly differ among the types of conduits except for standard polyester/PTFE [20].

In some cases, the surgical treatment may not be the most suitable option for the patient, but the conservative treatment (intravenous antibiotics, anti-inflammatory therapy, drainage or local irrigation) presents high-rate mortality (58 %–100 %) [13,21].

Considering the patient's age, comorbidities and severity of the

infection, our strategy was to combine antimicrobial therapy with ISR using silver-coated polyesters graft (Intergard Synergy).

There had been no consensus on the duration and type of antimicrobial therapy. A minimum of 2 weeks of intravenous therapy followed by an oral regimen is needed. The total duration of antimicrobial treatment can be from 3 months to 1 year, and even lifelong if surgery is not possible [10]. For tularemia cases, streptomycin, gentamicin, doxycycline, and ciprofloxacin are the first choice [11]. Our treatment was ciprofloxacin, and intravenous gentamicin for 2 weeks, followed by ciprofloxacin alone for 10 weeks. This management was successful and led to rapid recovery and no signs of recurrence so far.

4. Conclusions

We reported the first documented case of EVAR infection by *F. tularensis*. The diagnosis may be challenging because of nonspecific symptomatology and possibly negative CT or cultures. 99mTc-HMPAO-labeled WBC scintigraphy and PET scan were very helpful in identifying AEGI. The treatment of choice for AEGI is endograft removal and ISR, but conservative treatment may be acceptable for high-risk patients.

Ethics approval

All authors adhered to the guidelines for authorship mentioned in the "Guide for Authors" of the International Journal of Surgery Case Reports.

Ethics clearance was not necessary as it is not a study but a case report with a discussion according to a literature review.

Consent of patient

Written informed consent was obtained from the patient for publication of this case report and accompanying images in an anonymous way. A copy of the written consent is available for review by the Editor-in-Chief ofthis journal on request.

Consent for publication

All authors agreed with the content and gave explicit consent to submit this work.

CRediT authorship contribution statement

Manuscript writing: MK, AB, and BR; critical revision, manuscript approval, and agreement to be accountable: all authors; obtaining funding: BR and AB.

Declaration of competing interest

None.

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