Infected aortic endograft with an unusual microbe, *Burkholderia cepacia*

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ABSTRACT

With the growing use of endovascular aortic repair for aortic aneurysm pathology, multiple cases have been reported of associated endovascular graft infections. Explantation of the infected endograft and the revascularization procedure performed should be individualized with attention to the offending organism. We present the cases of two patients who underwent endovascular aortic repair with the same endograft and developed a graft infection with *Burkholderia cepacia*, a gram-negative organism with low virulence. Both endografts cultured *Burkholderia cepacia complex*; however, the organisms were genetically tested and found to be separate, unrelated strains. Both patients underwent successful explantation and revascularization procedures without any surgical-related complications to date. (J Vasc Surg Cases Innov Tech 2023;9:101295.)

Keywords: *Burkholderia cepacia*; Endograft infection

Each year in the United States, 45,000 abdominal aortic aneurysm (AAA) repairs are performed.[1] Endovascular aortic repair (EVAR) has grown as a treatment of ruptured and nonruptured AAAs and accounts for 74% of AAA repairs.[1] EVAR results in reduced morbidity and mortality compared with open AAA repair.[1] The complications of EVAR include endoleak, graft migration, component separation, and infection. Infection is a rare complication, reported in 0.4% to 3% of cases.[2] The recommendation is to treat aggressively with graft explantation and revascularization.[3] Traditionally, revascularization included extra-anatomic bypass to avoid placing a new graft in a potentially infected wound bed. In the present case series, we describe the cases of two patients with endograft infection due to *Burkholderia cepacia*, a gram-negative organism with low virulence. One patient underwent explantation and axillary femoral–femoral bypass. The second patient underwent explantation and anatomic reconstruction with a cryopreserved aorto-bi-iliac homograft. Both patients provided written informed consent for the report of their case details and imaging studies.

CASE REPORT

Patient 1. Patient 1 is a 77-year-old man with a 7.2-cm asymptomatic AAA. Elective EVAR was performed using Excluder endografts (W.L. Gore & Associates) without complications. Patient 1 had chronic myelogenous leukemia treated with imatinib. At 4 months after EVAR, he developed *B. cepacia* bacteremia due to cellulitis that was originally treated with empiric antibiotics before transitioning to meropenem for 7 days, followed by oral sulfamethoxazole-trimethoprim for 2 weeks.

At 8 months after EVAR, he developed abdominal and back pain. Computed tomography (CT) demonstrated a noncontrast-enhancing para-aortic fluid collection (Fig 1). The blood cultures were negative. Magnetic resonance imaging did not demonstrate osteomyelitis of the adjacent spine. He was initially managed with percutaneous drainage, culture, and intravenous antibiotics. Fluid culture resulted in *B. cepacia*, and the decision for explantation was made. The patient underwent explantation, which demonstrated purulent fluid in the aneurysm and retroperitoneum that cultured for *B. cepacia*. Extra-anatomic reconstruction was performed with axillary femoral–femoral bypass. The infected endograft was sonicated and cultured *B. cepacia*. No complications occurred postoperatively, and the patient was discharged home 7 days after his operation with a 6-week prescription for meropenem based on susceptibilities. He has been followed up for >2 years with repeat CT angiography (CTA) and has done well with no vascular complications.

Patient 2. Patient 2 is a 51-year-old woman who underwent urgent EVAR with Excluder devices (W.L. Gore & Associates) for a 4.5-cm AAA with inflammatory changes seen on CTA at an outside facility, 100 miles away. The inflammatory changes improved after EVAR. The patient developed a dental infection 4 months before EVAR but the culture data were not unavailable. She was transferred to our hospital 6 weeks later with concern for an infected endograft because of worsening back pain and a psoas abscess adjacent to the endograft on CT. The blood cultures were negative, and empiric antibiotics were started. The psoas abscess was aspirated and cultured *B. cepacia* (Figs 2-4).

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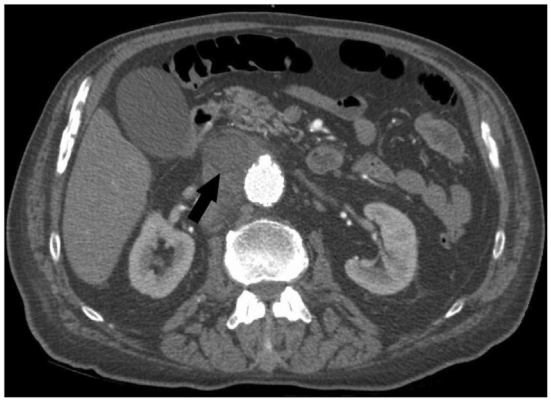

Fig 1. Axial reconstruction of computed tomography angiography (CTA) of patient A demonstrating the proximal endograft in the infrarenal segment and para-aortic abscess (*black arrow*).

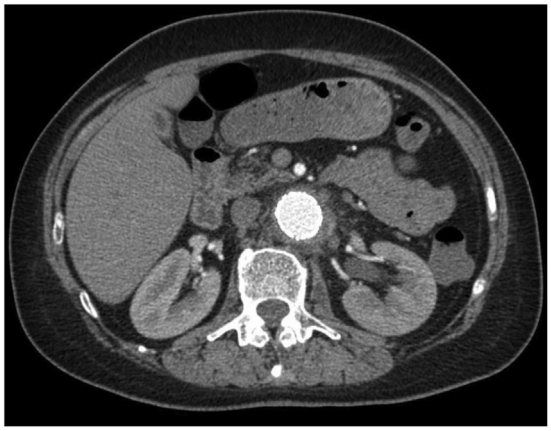

Fig 2. Axial reconstruction of computed tomography angiography (CTA) of patient B demonstrating inflammation in the periaortic space near the proximal endograft.

The patient also underwent a white blood cell-tagged scan, which showed uptake around the endograft and confirmed endograft infection.

Recognizing the short neck of the aneurysm below the renal arteries, she was taken to the operating room for explantation and anatomic aorto-bi-iliac homograft reconstruction with a vascularized omental flap. Inflammatory tissue surrounded the portion of the aorta with the infected graft, but no gross purulence was noted. Concern was present for an aortoenteric fistula; however, esophagogastroduodenoscopy and leak test results were negative. The endograft was sonicated and cultured *B. cepacia*. No major complications were encountered, and she was discharged home 7 days postoperatively with a 6-week prescription for meropenem based on susceptibilities. She has been seen in the clinic for >2 years with repeat CTA scans without any vascular issues.

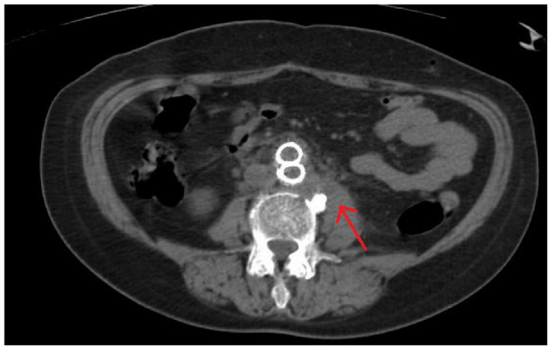

Fig 3. Axial reconstruction of computed tomography angiography (CTA) of patient B demonstrating a left psoas abscess adjacent to the endograft.

DISCUSSION

The endografts cultured *B. cepacia complex*. This is a group of ~20 gram-negative, catalase-producing, non–lactose-fermenting bacilli. This complex of bacteria was first described in 1950. Originally, *Burkholderia* was misclassified as *Pseudomonas* until 1992 when molecular analyses differentiated it. Modern bacterial cultures and polymerase chain reaction can differentiate *Burkholderia*; however, it can be misidentified because of its complexity.[4] They are nonvirulent compared with other gram-negative bacilli seen in endograft infections but can be resistant to multiple antibiotics. Often found in the soil, they are associated with severe pulmonary infections in patients with cystic fibrosis and have resulted in outbreaks due to contamination of over-the-counter products.[5] This is a rare organism for graft infection, because most infections are due to gram-positive cocci or are polymicrobial.[6] Because of the low virulence and rarity, it is important for both vascular surgeons and infectious disease physicians to realize this pathogen can cause indolent aortic graft infections.[4]

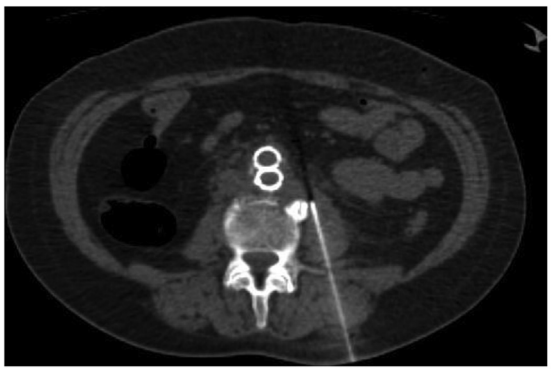

Fig 4. Axial reconstruction of computed tomography (CT) of patient B demonstrating interventional radiology aspiration.

Knowing the procedures occurred within 6 months of each other at separate facilities, we were concerned regarding an endograft source of the infection. The

device manufacturer was alerted, but no common source could be found. The organisms were genetically separate, unrelated strains with unique susceptibilities. One other case report demonstrated similar findings of an infected aortic graft with *Burkholderia pseudomallei*. That patient underwent explantation, creation of a neo-aorta iliac system with an autologous femoral vein, and prolonged intravenous and then oral antibiotics. They postulated that gardening in the perioperative setting led to seeding of the endograft.[7] *Burkholderia* is also found in water environments, including lakes, rivers, and drinking water.[8] Another possible source of contamination is ultrasound gel. A systematic review analyzed outbreaks of *Burkholderia* during a 30-year period and found 14 outbreaks associated with contaminated ultrasound gel.[9] Other case studies from Kazakhstan, India, Saudi Arabia, and Australia have also reported *Burkholderia* infections due to contaminated ultrasound gel.[10-13] At the time of our cases, ultrasound gel was not tested as a possible contaminant.

Reviewing the patient risk factors, patient 1 was relatively immunosuppressed because of chronic myelogenous leukemia. Moreover, he had developed *B. cepacia* bacteremia due to extremity cellulitis 4 months after EVAR, which was confirmed by three blood cultures. He was treated with 1 week of intravenous antibiotics, resulting in negative repeat blood cultures, followed by oral antibiotics. However, he developed an endograft infection ~3.5 months after completion of his antibiotic regimen. The index blood culture results differed from the more resistant strain cultured from the endograft, highlighting the antibiotic resistance this bacterium can develop.

Patient 2 did not have any comorbidities placing her at an increased risk of infection. The patient did report a dental abscess before EVAR, which is not typically caused by *Burkholderia*. It could not be excluded that her AAA became seeded at the time of her dental infection; however, no literature is available to support this concern.

CONCLUSIONS

Although endograft infections are rare, they are associated with high mortality and are a feared complication of EVAR. Causes of endograft infections are usually categorized by source: contamination during implantation, seeding from systemic infection, seeding from an adjacent infection, or enteric fistula.[3] Given the increasing use of EVAR, we expect that graft infections will be encountered more frequently by vascular surgeons. This case series highlights the cases of two patients with endografts infected with an uncommon, low-virulence, gram-negative rod treated with different surgical techniques for reconstruction. One patient received extra-anatomic reconstruction and the other received anatomic reconstruction with omental flap coverage. Both patients have done well clinically after completing antibiotic therapy. We suggest that both techniques are viable and reasonable options for surgical reconstruction after explantation of infected aortic endografts, even those infected with this low virulence gram-negative rod.

REFERENCES

1. Dua A, Kuy S, Lee CJ, Upchurch GR, Desai SS. Epidemiology of aortic aneurysm repair in the United States from 2000 to 2010. J Vasc Surg 2014;59:1512-7.
2. Daye D, Walker TG. Complications of endovascular aneurysm repair of the thoracic and abdominal aorta: evaluation and management. Cardiovasc Diagn Ther 2018;8:S138-56.
3. Cernohorsky P, Reijnen MM, Tielliu IF, van Sterkenburg SM, van den Dungen JJ, Zeebregts CJ. The relevance of aortic endograft prosthetic infection. J Vasc Surg 2011;54:327-33.
4. Lipuma JJ. Burkholderia cepacia complex as human pathogens. J Nematol 2003;35:212-7.
5. Sousa SA, Ramos CG, Leitão JH. Burkholderia cepacia complex: emerging multihost pathogens equipped with a wide range of virulence factors and determinants. Int J Microbiol 2011;2011:607575.
6. Murphy EH1, Szeto WY, Herdrich BJ, et al. The management of endograft infections following endovascular thoracic and abdominal aneurysm repair. J Vasc Surg 2013;58:1179-85.
7. Cheok S, Gan LSC, Chung SJ, Ch'ng JK. Aortic endograft infection secondary to Burkholderia pseudomallei: a case report and review of the literature. J Vasc Surg Cases Innov Tech 2021;7:421-4.
8. Tavares M, Kozak M, Balola A, Sá-Correia I. *Burkholderia cepacia* complex bacteria: a feared contamination risk in water-based pharmaceutical products. Clin Microbiol Rev 2020;33:e00139-219.
9. Angrup A, Kanaujia R, Biswal M, Ray P. Systematic review of ultrasound gel associated Burkholderia cepacia complex outbreaks: clinical presentation, sources and control of outbreak. Am J Infect Control 2022.
10. Viderman D, Khudaibergenova M, Kemaikin V, Zhumadilov A, Poddighe D. Outbreak of catheter-related Burkholderia cepacia sepsis acquired from contaminated ultrasonography gel: the importance of strengthening hospital infection control measures in low resourced settings. Infez Med 2020;28:551-7.
11. Solaimalai D, Devanga Ragupathi NK, Ranjini K, et al. Ultrasound gel as a source of hospital outbreaks: Indian experience and literature review. Indian J Med Microbiol 2019;37:263-7.
12. Abdelfattah R, Al-Jumaah S, Al-Qahtani A, Al-Thawadi S, Barron I, Al-Mofada S. Outbreak of Burkholderia cepacia bacteraemia in a tertiary care centre due to contaminated ultrasound probe gel. J Hosp Infect 2018;98:289-94.
13. Shaban RZ, Maloney S, Gerrard J, et al. Outbreak of health care-associated Burkholderia cenocepacia bacteremia and infection attributed to contaminated sterile gel used for central line insertion under ultrasound guidance and other procedures. Am J Infect Control 2017;45:954-8.

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