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Case Report

A Case of Postthoracic Endovascular Aortic Repair Stent-Graft Infection Caused by *Campylobacter insulaenigrae*

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An 81-year-old Japanese man who had distal aortic arch dilatation at age 77 had thoracic endovascular aortic surgery. The patient developed a fever and was taken to the hospital. Reduced diffusion in the descending aortic wall along the stent graft was discovered using whole-body diffusion-weighted imaging with background body signal suppression, and stent-graft infection was identified. The 16S ribosomal RNA gene analysis and blood culture results identified *Campylobacter insulaenigrae* as the etiological bacterial species. The patient was released from the hospital after 6 weeks of antibiotic treatment since the swelling and inflammatory response had decreased.

Keywords: *Campylobacter insulaenigrae*, stent-graft infection

Introduction

The bacterium *Campylobacter insulaenigrae* was discovered in marine mammals,[1] and human infection is extremely uncommon. Only two human infection cases have ever been documented globally, and there have been no instances of cardiovascular infections.[2,3] This report details an incredibly unusual instance of *C. insulaenigrae* thoracic aortic stent-graft infection that occurred after our hospital's thoracic endovascular aortic repair (TEVAR) procedure.

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Case Report

An 81-year-old Japanese man with Stanford type A acute aortic dissection had blood vascular prosthesis implantation in the aortic arch in 2014 (at age 72 years). He had TEVAR in 2018 (at age of 77) for a persistent dissecting aortic aneurysm in the distal arch. After the procedure, the patient underwent follow-up observation after the postoperative evaluation revealed no endoleaks. The patient became ill in April 2022 with a fever and was admitted to the hospital to identify the cause. His medical history includes chronic renal failure, hypertension, and angina pectoris, all of which required percutaneous coronary intervention.

The patient was awake and cognizant when admitted. The vital signs were as follows: body temperature, 38.0°C; blood pressure, 133/51 mmHg; pulse rate, 89/min (regular pulse); and SpO_2, 95% (room air). Blood testing showed a high C-reactive protein level of 31.6 mg/dL and a high white blood cell count of $12.0 \times 10^3/\mu L$. Cefazolin (1 g/8 h) was started as soon as the patient was admitted. A dilatation in the descending aorta was discovered by contrast-enhanced computed tomography (CT) on day 3 of the patient's hospitalization (**Figs. 1a** and **1b**). On day 5, the presence of gram-negative bacillus (spirillum) was detected in a blood culture sample collected on admission, suggesting possible infection with either a *Campylobacter* or *Helicobacter*. This prompted us to change the antibiotic used to meropenem (1 g/12 h). On day 8, a relapse of inflammatory reaction and pyrexia occurred, and the regimen was changed to cefazolin (1 g/8 h) plus meropenem (0.5 g/12 h) plus pazucross (0.5 g/12 h). On day 9, whole-body diffusion-weighted imaging with background body signal suppression showed decreased diffusion in the descending aortic wall along the stent graft, leading to a diagnosis of stent-graft infection (**Fig. 1c**). Given the patient's advanced age, a therapeutic regimen was formulated to administer conservation therapy with antibiotics, and surgery did not result in improvement. On day 15, the findings of the blood culture initiated on admission re-

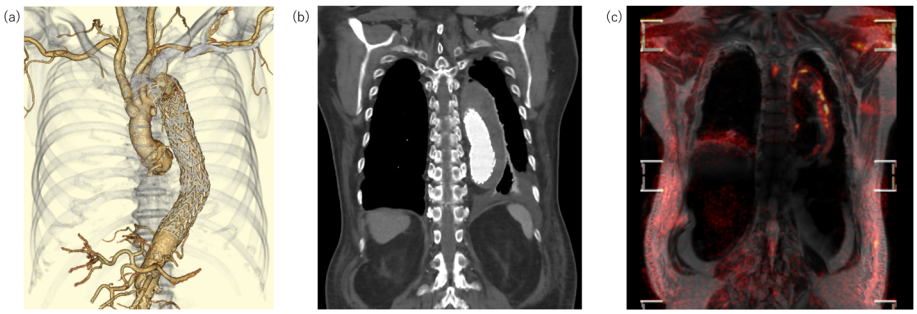

Fig. 1 (**a**) Three-dimensional computed tomography at 3 days after admission. (**b**) Enhanced computed tomography at 3 days after admission. (**c**) Whole-body diffusion-weighted imaging with background body signal suppression at 9 days after admission.

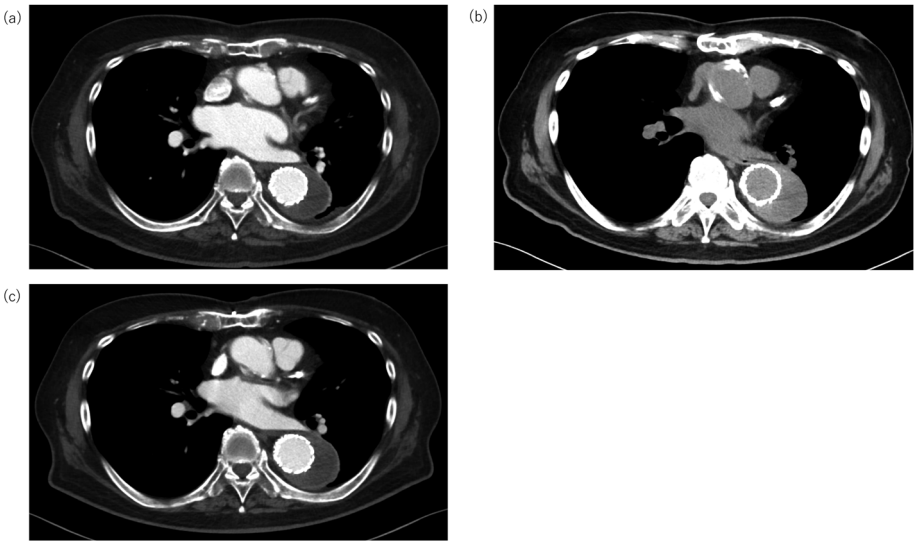

Fig. 2 Computed tomography showed a descending aortic aneurysm (Th7/Th8 level). (**a**) Enhanced computed tomography at 3 days after admission (maximum diameter was 61 mm). (**b**) Computed tomography at 32 days after admission (maximum diameter was 63 mm). (**c**) Enhanced computed tomography at 54 days after admission (maximum diameter was 61 mm).

vealed the responsible pathogen as *Campylobacter*; however, the bacterial species could not be identified. To prevent tolerance to cephems and quinolones, the antibiotics used were changed to meropenem (1 g/8 h) plus arbekacin (200 mg/48 h). Another hospital was commissioned to perform 16S ribosomal RNA gene analysis, which identified the causative bacterium as *C. insulaenigrae* on day 18 (two sets of blood cultures were collected on admission). Tolerance to cephems and quinolones was also noted. The patient had no history of contact with marine mammals or ingestion of supplements derived from marine mammals. On day 32, CT showed an expansion in the diameter of the descending aorta (**Figs. 2a** and **2b**). Antibiotics were administered for 6 weeks after the prescribed antibiotics were changed to meropenem and arbekacin. However, since the patient experienced stomach discomfort and loss of appetite, the antibiotics were stopped on day 46, and no pyrexia was noticed thereafter. On day 54, contrast-enhanced CT revealed shrinkage of the descending aorta (**Fig. 2c**). On day 58, blood testing revealed a high white blood cell count of $8.2 \times 10^3/\mu L$ and high C-reactive protein level of 6.7 mg/dL. On day 57, whole-body diffusion-weighted images with background body signal suppression showed that there was no longer a reduction in diffusion in the descending aortic wall along the stent graft (**Fig. 3a**). On day 59, the patient was released from hospital. At 4 months after discharge, the diameter of the descending aorta was found to have further shrunken, and no recurrence of infection was noted (**Fig. 3b**). At 9 months after discharge, blood testing showed a high white blood cell count of $7.8 \times 10^3/\mu L$ and high C-reactive protein level of 1.2 mg/dL.

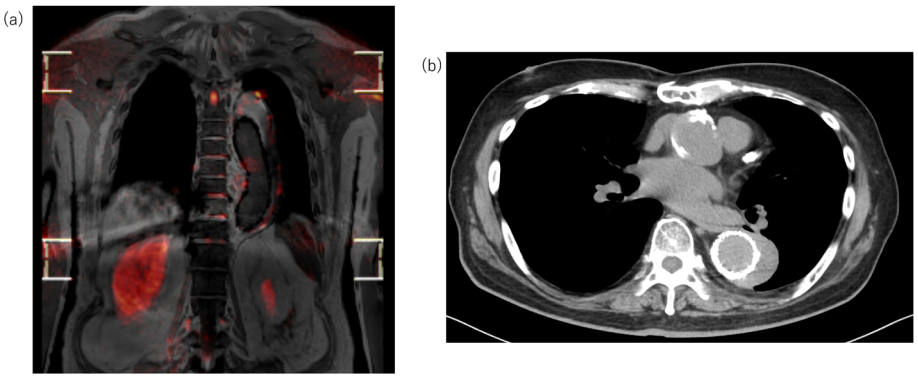

Fig. 3 (**a**) Whole-body diffusion-weighted imaging with background body signal suppression at 57 days after admission. (**b**) Computed tomography at 4 months after discharge (maximum diameter was 46 mm).

Discussion

We recently encountered a case of thoracic aortic stent-graft infection caused by *C. insulaenigrae*. Only two cases of human infection with *C. insulaenigrae* have been previously reported: infective gastroenteritis reported by Chua et al. and meningitis reported by Kyotani et al. The present report constitutes a third case and the first report of infection in the cardiovascular region.[2,3] While *C. insulaenigrae* is a bacterium identified from marine mammals,[1] this patient had no history of exposure with marine mammals or ingestion of supplements sourced from marine mammals. Therefore, the portal of entry remained unknown as with the previous two cases.[2,3] Given the extreme rarity of this bacterium, we think continued accumulation of further case studies is important.

Recently, as stent-graft implantation in the thoracic aorta has become widespread, the incidence of thoracic aortic stent-graft infection has been increasing. With the incidence rate ranging from 1.53% to 4.77% and the occurrence rate of complications from 25% to 75%, there has been growing concern.[4] To treat stent-graft infection, the preferred method is to completely remove the stent graft and close the infection site if debridement and fistulas are noted in the infectious lesion.[5–7] However, many cases require stent-graft treatment due to the inadequacy of the standard blood vascular prosthesis implantation modality, and in such cases, the risk of surgery increased.[8,9] Likewise, since the present case did not develop an abscess cavity, conservation therapy using antibiotics was first selected, and a surgical procedure would be implemented if infection control could not be achieved. Shukuzawa et al. showed the effectiveness of conservation therapy against abdominal aortic stent-graft infection.[10] Similarly, conservation therapy may also be effective against thoracic aortic stent-graft infection in cases without any fistula or abscess cavity. In this instance, the patient achieved remission by conservation therapy using antibiotics. However, if infection relapses in the future, surgical treatment may be required in light of the possibility that the causative bacterium has acquired multiple drug resistance.

Imaging diagnosis of prosthetic vascular infection is typically made using a CT scan, which shows fluid retention and gas images around the prosthesis.[11] Positron emission tomography (PET)-CT is also an effective diagnostic tool.[12] In this instance, CT could not confirm the diagnosis and PET-CT was unavailable in the hospital; therefore, whole-body diffusion-weighted magnetic resonance imaging was performed, which shows the areas with decreased diffusion motion of water molecules that are associated with cellular edema and necrosis as high-signal areas. Since reduction of water molecule diffusion is seen in acute inflammatory areas such as tumors, abscesses, infections, and the lesion site and its extent can be determined.[13] Although its utility has not been reported in the field of cardiovascular surgery, its usefulness in the diagnosis of multi-organ diseases such as brain, liver, and kidney has been reported,[14–16] and we believe it can aid in diagnosing artificial vascular infection.

Conclusion

We encountered a case of thoracic aortic stent-graft infection caused by *C. insulaenigrae*. Given the exceptional rarity of this bacterium, further accumulation of case studies is warranted.

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Informed Consent

Informed consent was obtained in August 2018.

Disclosure Statement

All authors declare that there is no conflict of interest regarding the publication of this paper.

Author Contributions

Data collection: all authors
Writing: KY
Revision: KY
Critical review and revision: all authors
Final approval of the article: all authors
Accountability for all aspects of the work: all authors

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