ESC European Society of Cardiology

European Heart Journal - Case Reports (2020) **4**, 1–5
doi:10.1093/ehjcr/ytaa081

CASE REPORT
Congenital heart disease

Infectious stentitis after treatment of coarctation of the aorta: a case report

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Received 17 October 2019; first decision 18 November 2019; accepted 17 March 2020; online publish-ahead-of-print 3 May 2020

Background Aortitis is a rare condition that can be caused by inflammatory or infectious aetiologies. The clinical presentation of aortitis includes a heterogeneous range of symptoms and clinical signs.

Case summary We present a 53-year-old man whose medical history included presence of a ventricular septal defect, a bicuspid aortic valve, and coarctation of the aorta. The coarctation was treated with percutaneous stent implantation. One and a half years later, he presented to our hospital with complaints of fatigue, night sweats, and shivers. Physical examination revealed a fever, tachycardia, and hypertension. Imaging studies showed no signs of endocarditis. Positron emission tomography–computed tomography (PET–CT) showed an increase in ^{18}F-fluorodeoxyglucose uptake at the distal end of the stent in the descending aorta. Blood cultures revealed a *Streptococcus gordonii* and antibiotic treatment was adjusted accordingly. The patients' functional status improved quickly, the fever resolved, and the laboratory markers of inflammation returned to normal.

Discussion Aortitis is extremely rare after stent implantation. Risk factors for aortitis include congenital vascular malformation and stent implantation. Computed tomography is currently the imaging study of choice for aortitis, while PET–CT seems ideal for identification of stent infection. Mortality associated with infectious aortitis ranges from 21% to 44%, with generally higher mortality if managed with antibiotics alone. The differential diagnosis of stent infection should be taken into account in patients presenting with fever and chills after previous stent procedures.

Keywords Infection • Coarctation of the aorta • Stenting • Case report

Learning points

- The differential diagnosis of stent infection should be taken into account in patients presenting with fever and chills after previous stent procedures.
- Surgery should be considered as treatment in urgent cases, especially when aneurysm formation is present.

Introduction

Surgical repair or catheter-based stenting is recommended for adult patients with hypertension and significant coarctation of the aorta (CoA).[1,2] Various complications have been described afterwards, including recoarctation, aneurysm formation, and dissection. Long-term outcomes have been provided by various research groups, ranging from 99% at 30 years of age to 65% survival at 70 years of

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Handling Editor: Monika Arzanauskaite

Peer-reviewers: Christoph Sinning and Julien Adjedj

Compliance Editor: Amir Aziz

Supplementary Material Editor: Vassiolios Parisis Memtsas

age.[3–5] Event-free survival have been reported from 96.7% 10 years after initial treatment to 53% at 70 years of age.[3–5]

Aortitis is a rare condition that can be caused by inflammatory or infectious aetiologies. The clinical presentation of aortitis includes a heterogeneous range of symptoms and clinical signs, largely determined by the underlying cause, the location of aortic wall thickening and the presence of coexisting arteries at the side of the aortitis.[6] Here, we present a case of infectious aortitis in the presence of a bare-metal stent used for treatment of coarctation of the aorta. Written informed consent by the patient was obtained.

Timeline

Case	A 53-year-old man with a history of endocarditis, a ventricular septal defect, and coarctation of the aorta treated by percutaneous stent implantation, presented with fatigue, night sweats, and fever. Blood cultures were taken and antibiotic treatment was initiated.
Day 1	Transthoracic echocardiography showed no signs of endocarditis.
Day 3	Transoesophageal echocardiography showed no signs of endocarditis. Blood cultures identified *Streptococcus gordonii*. Antibiotic treatment was switched accordingly.
Day 7	Positron emission tomography–computed tomography (PET–CT) scan showed an increase in ^{18}F-fluorodeoxyglucose (FDG) uptake at the distal end of the stent in the descending aorta.
Day 24	Clinical improvement, no more complaints or fever. Markers of inflammation returned towards normal. Repeat PET-CT: normalization of the FDG uptake at the distal stent end. Discharge home.
Day 49	Completion of intravenously administered antibiotic treatment.

Case presentation

A 53-year-old man was referred to our hospital for a second opinion because of the suspicion of infection of an aortic stent. His medical history included presence of a small, restrictive muscular ventricular septal defect (VSD), a bicuspid aortic valve (BAV) of the right–left coronary cusps type, and coarctation of the aorta. More than 30 years before presentation, he had a sepsis with a *Staphylococcus aureus* and was treated as if it were endocarditis. In 2016, the coarctation was treated percutaneously by uncomplicated implantation of a Cheatham-Platinum/Iridium CP stent (NuMed Inc. Hopkinton, NY, USA) because of persistent hypertension. A non-covered stent was used to allow unrestricted perfusion of the lusoric artery through the stent struts. Approximately one and a half years later, he presented to the referring hospital with fatigue, night sweats, and shivers that persisted for several weeks. Physical examination revealed fever up to 40°C combined with a blood pressure of 153/78 mm of mercury,

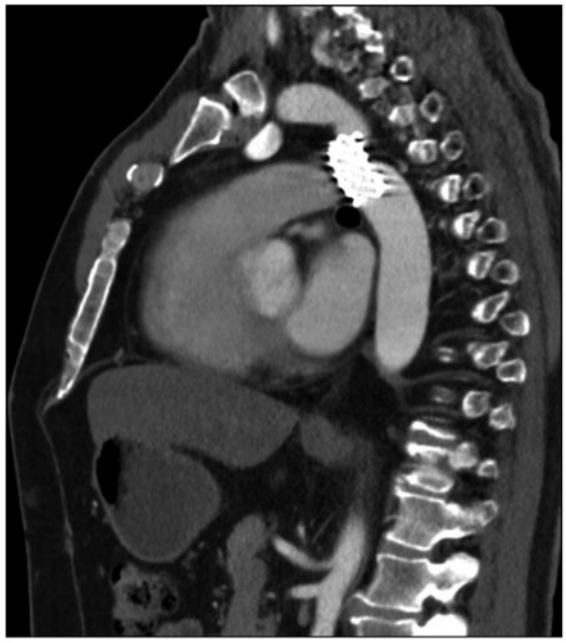

Figure 1 The computed tomography scan showing the CP stent in the aortic arch/descending aorta one day after implantation without any signs of inflammation.

a tachycardia of 106 b.p.m., and a known systolic murmur on the aortic valve. The electrocardiogram showed a sinus tachycardia including a first-degree atrioventricular block. Laboratory results included leucocytes 11.1 × 10e^9/L (reference range 4.0–10.0 × 10e^9/L), C-reactive protein 66 mg/L (0–10 mg/L), haemoglobin 8.2 mmol/L (8.6–10.7 mmol/L). He was admitted with the working diagnosis of endocarditis and after taking blood cultures antibiotic treatment consisting of ceftriaxone was initiated. Transthoracic echocardiography showed no proof of endocarditis and therefore, transoesophageal echocardiography was performed: the BAV functioned well, the peak velocity was 1.8 m/s, no aortic valve regurgitation was seen, and no vegetation or other characteristics of endocarditis were found. The entry point was extensively searched for but could not be identified. Blood cultures revealed a *Streptococcus gordonii* and antibiotic treatment was adjusted accordingly to penicillin 12 million units per 24 h. A computed tomography (CT) scan was performed and showed no vegetation or thrombus of the BAV, no wall thickening, aneurysm, abscess, or other signs of inflammation (*Figure 1*). A positron emission tomography (PET)–CT scan with ^{18}F-fluorodeoxyglucose (FDG) showed an increase in FDG uptake at the distal end of the stent in the descending aorta (*Figure 2*). Hereafter, antibiotic treatment was switched again to ceftriaxone once daily 2000 mg and continued for 6 weeks, in accordance with treatment of endocarditis with a prosthetic valve.[7] Clinically the patients' functional status improved quickly, the fever resolved and the laboratory markers of inflammation returned to normal. Control PET–CT scan a week after treatment showed normalization of FDG uptake (*Figure 3*). The patient was sent home where he completed the antibiotic treatment. During outpatient follow-up, he had no fever and control blood cultures

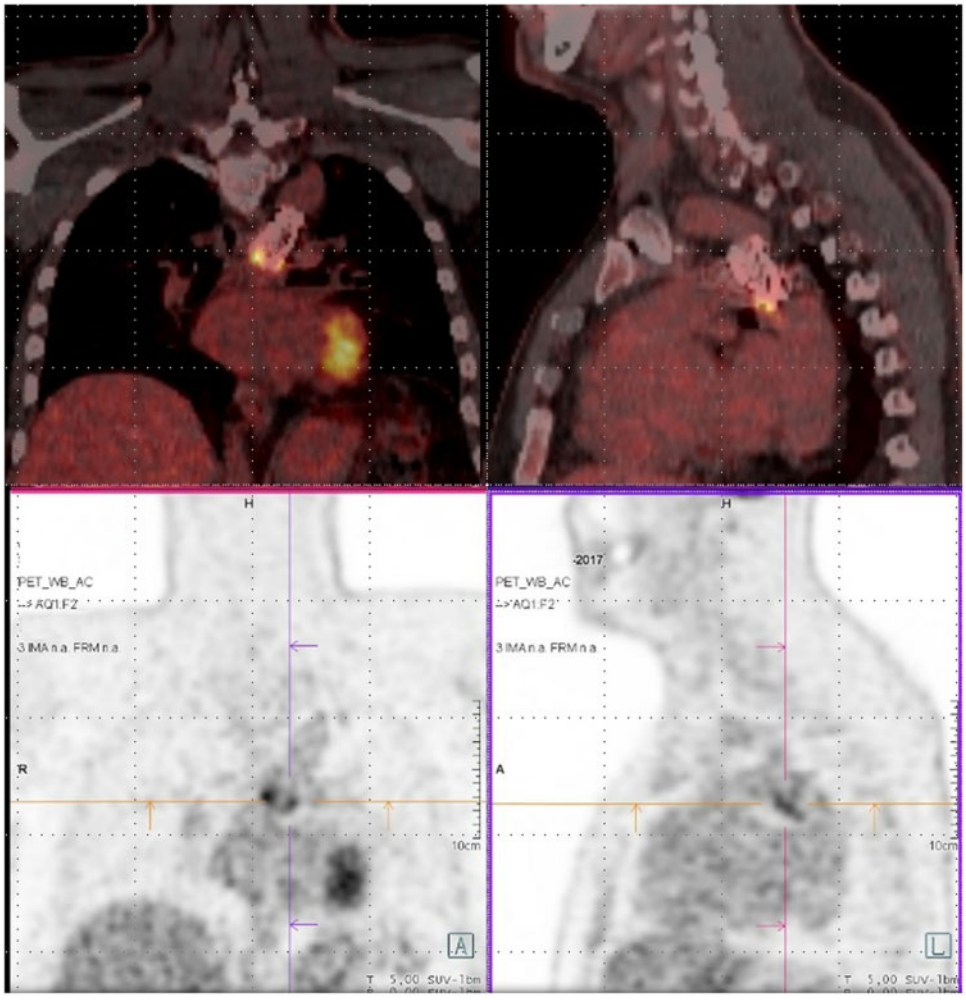

Figure 2 The positron emission tomography–computed tomography scan with ^{18}F-fluorodeoxyglucose showing increased ^{18}F-fluorodeoxyglucose uptake at the distal end of the stent in the proximal descending aorta.

remained negative. At last follow-up, August 2019, he was free of complaints.

Discussion

We present a rare case of aortitis after stenting of a CoA. Patients with congenital heart disease have an increased risk of developing infective endocarditis and this risk is related to the type of defect encountered.[1,2,8] Beforehand, our patient could have an infection related to the BAV, the VSD, and aortic stent. Transthoracic nor transoesophageal echocardiography showed signs of endocarditis and therefore we chose for a PET–CT scan that is considered an important supplementary method for patients with suspected infective endocarditis.[7] By doing so, we identified infection of the aortic stent and no abnormalities at the side of the BAV or VSD were seen.

Percutaneous stent implantation is a standard treatment of CoA in adulthood. Aortitis is extremely rare after stent implantation. As we stated earlier, in the current case, a PET–CT scan could demonstrate situation of the infectious substrate at the stent and its infectious activity dissolved after antibiotic therapy. Under normal conditions, the aorta is resistant to infection, but risk factors such as congenital vascular malformation[9] or stent implantation[6,10] have been identified for the development of infectious aortitis. It seems reasonable that the vessel trauma caused by stent implantation may be a predilection area for aortitis.

Both transoesophageal echocardiography as well as 99m-TC labelled white blood cell scintigraphy have been used to diagnose aortitis and aortic prosthetic stent infection.[11–14] Computed tomography with contrast is currently the imaging study of choice for aortitis,[9] while a PET–CT seems to be an ideal medium for identification of stent infection.[15]

The most common pathogens causing infectious aortitis have been identified, being Salmonella species, *S. aureus*, streptococcal species, Gram-negative bacilli other than Salmonella and fungi.[9] Mechanisms of infection include haematogenous spread, contiguous seeding from adjacent infection, and traumatic or iatrogenic inoculation.[16] *Streptococcus gordonii*, which was found in our patient, is a normal inhabitant of the

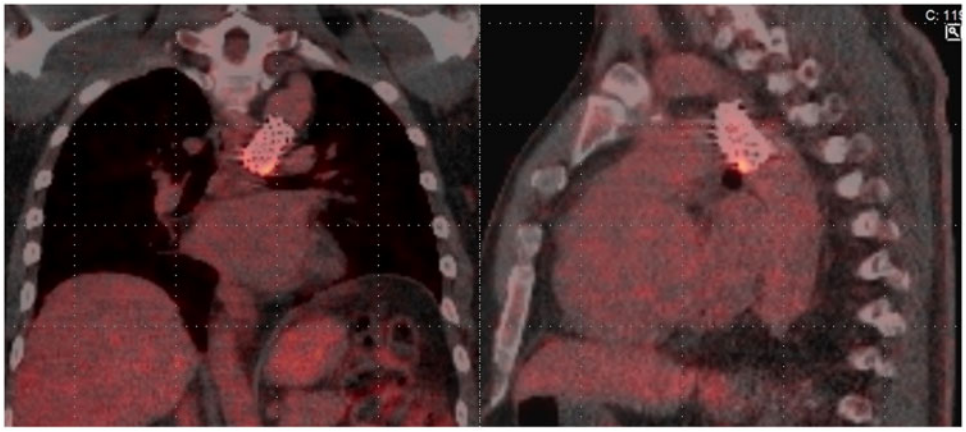

Figure 3 Control positron emission tomography–computed tomography at follow-up scan showing normalization of ^{18}F-fluorodeoxyglucose uptake.

human oral cavity. It is regarded as one of the main causative agent in the development of subacute bacterial endocarditis.

The CP stent is widely used for the treatment of large vessel stenosis in various congenital heart diseases. In a recent publication, no stentitis was reported during a 12-month follow-up.[17] In general, intravascular bare metal stent infections are a rare but potentially serious complication as Bosman *et al.*[15] found in their review of literature. Mortality associated with infectious aortitis ranges from 21% to 44%, with generally higher mortality if managed with antibiotics alone.[9] Mlynski *et al.*[18] reported a case of pneumococcal aortitis secondary to endovascular bare-metal stent infection 1 year after stent implantation that was complicated by aortic rupture. Urgent surgical treatment led to a favourable outcome. This underlines the importance of considering surgery in urgent cases, especially when aneurysm formation is present.

In conclusion, infectious aortitis may rarely occur after stent implantation for treatment of coarctation of the aorta. The differential diagnosis of stent infection should be taken into account in patients presenting with fever and chills after previous stent procedures. In our case, it has been treated successfully by medical therapy alone. Surgery has to be considered in urgent cases of aortitis.

Lead author biography

Heleen B. van der Zwaan currently works as a cardiologist at the University Medical Center in Utrecht where she did specialization in adult congenital heart disease. She completed her thesis on right ventricular function assessment by three-dimensional echocardiography in patients with various congenital heart diseases at the Erasmus Medical Center in Rotterdam. She is interested in cardiac imaging and treatment of advanced heart failure therapy.

Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as Supplementary data.

Consent: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: none declared.

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