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CASE REPORT
Arrhythmias / Electrophysiology

Pacemaker lead rupture in a patient with subacute endocarditis: a case report

Lorenzo Caratti di Lanzacco [1] and Aurélien Wauters [1,2,*]

[1]Cardiology Department, Cliniques Universitaires St-Luc, Université Catholique de Louvain, Av. Hippocrate 10, 1200 Brussels, Belgium; and [2]Cardiology Department, Clinique St-Pierre, Avenue Reine Fabiola 9, 1340 Ottignies-Louvain-La-Neuve, Belgium

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Background Cardiac implantable electronic device (CIED)-related infections are associated with severe morbidity and mortality. Few cases have previously documented both lead endocarditis and lead rupture simultaneously.

Case summary We describe the case of a 73-year-old man with a dual-chamber pacemaker presenting with subacute endocarditis and recurrent cholangitis. A few months prior, the patient was diagnosed with localized colon cancer and *Streptococcus sanguinis* lead endocarditis based on nuclear imaging. He was given prolonged antibiotic therapy and lead explantation was to be performed after sigmoidectomy. During the following weeks, his condition worsened and he was readmitted for biliary sepsis. A chest X-ray revealed, incidentally, a complete ventricular lead rupture. Pacemaker electrogram showed ventricular undersensing, loss of ventricular capture, and high impedance. As his health declined, removal of the pacemaker was deemed unreasonable and the patient died of biliary sepsis in the next few weeks.

Discussion We describe the case of an asymptomatic intracardiac lead fracture in the setting of colon cancer and a medically managed *Streptococcus* lead infection. As this complication occurred during lead infection, bacterial damage may have weakened the lead over time. As illustrated by the patient's outcomes, long-term antibiotic therapy should only be used in cases unsuitable for device removal. Complete hardware removal remains the first-line therapy in patients with CIED-related infections.

Keywords Pacemaker • Rupture • Fracture • Pacemaker lead endocarditis • *Streptococcus sanguinis* • Case report

ESC Curriculum 4.11 Endocarditis • 5.9 Pacemakers

Learning points

- *Streptococcus sanguinis* species account for a high percentage of cardiac implantable electronic device (CIED)-related infections in cases of streptococcaemia. Clinicians should look for gastrointestinal neoplasm in cases of streptococcaemia.
- Microbial abrasion may contribute to lead fracturing in CIED-related endocarditis.
- Complete hardware removal is the first-line therapy for patients with CIED-related infection. Long-term suppressive antibiotic therapy bears higher morbidity–mortality and should only be considered in selected cases.

Introduction

Infections of cardiac implantable electronic devices[1] (CIEDs; pacemakers, implantable cardioverter-defibrillators, and cardiac resynchronization devices) are associated with high morbidity and mortality. Due to the ongoing rise in CIED implantations and ageing of the population, reports reveal an increase in CIED-related infections with an incidence around 1.9 per 1000 device-years[2] in spite of

* Corresponding author. Tel: +32 10 43 73 47, Email: aurelien.wauters@cspo.be
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the current technological progress, experienced centres, and systematic use of preoperative antibiotic prophylaxis.

Cardiac device-related endocarditis refers to an infection of the electrode leads, valve leaflets, and/or endocardium. Diagnosis relies primarily on blood cultures and echocardiography although requiring nuclear imaging in difficult situations.

The treatment of CIED-related infections is always complex, long-lasting and associated with severe complications.[2,3] We report here a rare case of lead dysfunction in the context of lead endocarditis.

Timeline

Time	Events
December 2009	Dual-chamber pacing and sensing system pacemaker implantation for paroxysmal third-degree atrioventricular block.
Late April 2017	Pacemaker box replacement due to battery end-of-life.
Early May 2020	Admitted for subacute endocarditis with: • *Streptococcus sanguinis* bacteraemia. • Right atrium vegetation around one of the leads. • Discovery of localized colon cancer on positron emission tomography–computed tomography (PET-CT). • Discharged home with long-term antibiotic therapy. Developed acute cholangitis during his stay: • Endoscopic retrograde cholangiopancreatography showed main bile duct lithiasis and hilar bile duct stenosis (biopsied). • Improvement following intravenous antibiotics and endoscopic stones extraction.
Late May	Normal pacemaker function in routine assessment.
Mid-June	Laparoscopic sigmoidectomy for localized colon cancer.
Late August	Admitted in the intensive care unit for biliary septic shock: • Treated with vasopressors, intravenous (IV) antibiotics, and endoscopic biliary drainage.
Early September	Transferred to gastroenterology ward once stabilized: • Presented with cholangitis recurrence due to drains obstruction. Improved on IV antibiotics and after biliary drain replacement.
Late October	Critically ill patient following biliary sepsis recurrence: • Decision not to replace the biliary drains. Antibiotic management alone. • Lead explantation is abandoned and the patient is discharged home with a lifelong antibiotic.
November 2020	Patient died from biliary sepsis.

Case presentation

A 72-year-old man with a prepectoral dual-chamber pacemaker implanted transvenously in 2009 (Zephyr XL DR, St-Jude Medical, St-Paul, MN, USA) for paroxysmal complete atrioventricular (AV) block and a box change in 2017 (Accolade MRI L311, Boston Scientific Corporation, MN, USA) programmed in dual-chamber pacing and sensing system was admitted in May 2020 for recurrent episodes of chills and confusion. The patient was afebrile, non-septic and had compensated heart failure at that time. His chills and confusion episodes developed 3 months prior and he received valproic acid for erroneously suspected epileptic seizures. Two strokes (2000 and 2009) left him with phasic disorder, cognitive impairment, and right hemiparesis. He had no past history of parenteral drug use.

After his hospital admission, blood samples demonstrated cholestasis and blood cultures isolated penicillin-sensitive *Streptococcus sanguinis*. Serum alkalin phosphatase reached 558 U/L [normal value (NV) < 169], total bilirubin was 2.3 mg/dL (NV < 1.2) with direct bilirubin at 1.6 mg/dL (NV < 0.5), alanine transaminase 51 U/L (NV < 44), absolute neutrophil count 23.840/mm^3 (NV 1.500–7.000), and C-reactive protein 89 mg/L (NV < 5). Chest computed tomography (CT) showed multiple pulmonary embolisms and transoesophageal echocardiography revealed a 41 × 33 × 15 mm mobile vegetation located on the intracardiac portion of the ventricular lead (*Figure 1*) (Zephyr 1948, St-Jude Medical, MN, USA). During his stay, the patient developed signs of biliary sepsis. Endoscopic retrograde cholangiopancreatography revealed common bile duct lithiasis and hilar bile duct stenosis. Intravenous antibiotics [Cefuroxime 1.500 mg/every 8 h (q8h) plus Metronidazole 500 mg/q8h] and endoscopical stones extraction improved his condition. Brush and forceps biopsies were performed in search for a bile duct malignancy. Lastly,

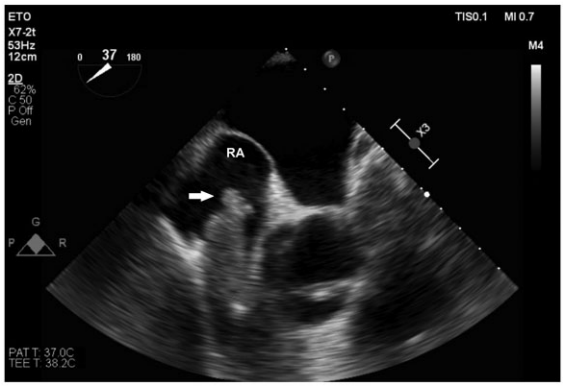

Figure 1 Transoesophageal echocardiograghy shows a 41 × 33 × 15 mm vegetation (arrow), in the right atrium, entrapping the ventricular pacemaker lead. RA, right atrium.

a localized sigmoid cancer was identified on ^{18}Fluorodesoxyglucose (^{18}F-FDG) positron emission tomography–CT (PET/CT) and he underwent sigmoidectomy the following month. Endovascular lead explantation was scheduled after rehabilitation. He was prescribed an oral first-generation cephalosporin (cephalexin 500 mg b.i.d.) as prolonged antibiotic therapy whilst awaiting lead removal.

During the next months, the patient's condition worsened as he was readmitted for recurrent cholangitis despite repetitive antibiotic therapies and endoscopic biliary drainages. Cholangiocarcinoma was suspected regardless of the negative cytological and histological results. Five months after the endocarditis' diagnosis, rest electrocardiogram (*Figure 2A*) demonstrated sinus rhythm with normal AV delay, ventricular pacing spike with loss of ventricular capture and ventricular undersensing. Chest X-ray (*Figure 3*) revealed a complete intracardiac ventricular lead fracture, which was absent 40 days prior. Pacemaker interrogation reported ventricular undersensing, loss of capture, and high impedance (>3000 ohms) (*Figure 2B*). A routine pacemaker assessment performed 4 months prior showed normal function and low rate of ventricular pacing (<1%) with a gradual decrease in ventricular pacing impedance (*Figure 2C*). Additionally, another ^{18}F-FDG PET/CT scan (*Figure 4*) demonstrated an increased uptake of ^{18}F-FDG in both the intrathoracic and extrathoracic portions of the pacemaker. Considering the extension of the infectious diseases and the comorbidities of the patient (palliative approach for suspected cholangiocarcinoma, two strokes and cognitive impairment), complete hardware removal was not a reasonable option. Instead, he was discharged home on lifelong antibiotic therapy (cephalexin).

The patient became critically ill after his third hospital stay for cholangitis. With the family's consent, he was later transferred to a palliative care unit where he passed away due to biliary sepsis.

Discussion

This case describes an association between lead endocarditis and endocardial lead rupture in the setting of a streptococcal infection. In comparison with staphylococci responsible for up to 80% of all CIED infections, other Gram positive cocci only account for 4% of cases.[3] The risk of infective endocarditis (IE) is highly species-dependent in the event of streptococcaemia.[4] *Streptococcus sanguinis* bloodstream infections result more often in IE (34.6%) than other streptococci (mean 7.1%).[5] Even if the association between *Streptococcus gallolyticus* (*Streptococcus bovis*) and IE is better established, *S. sanguinis* is a common cause of streptococcaemia in patients with underlying malignancies.[6] Up to 26.8% of patients with *Streptococcus sanguis* (*S. sanguinis*) bactaeraemia have cancer.[7] Comparatively, the prevalence of colon cancer ranges between 25% and 80% with *S. bovis* blood infections.[6] By disrupting intestinal mucosa, gastrointestinal neoplasms facilitate bacterial translocation into the blood. In our patient's case, the fact that both diagnoses of sigmoid cancer and *S. sanguinis* bacteraemia coincided indicates a relation between his colon cancer and the *S. sanguinis* IE.

The diagnosis of CIED-related endocarditis can be challenging. Nuclear imaging (^{18}F-FDG PET/CT scan and radiolabelled leucocyte scintigraphy) improves the sensitivity of the modified Duke criteria up to 80–90% for CIED-related infections.[8] Notably, PET/CT plays a key role when confronted with an intracardiac mass on echocardiography to differentiate vegetations from thrombus.[9] Nuclear imaging confirmed our patient's diagnosis of lead endocarditis.

The annual incidence of pacemaker lead fractures has been reported to be between 0.1% and 4.2%.[10] They commonly occur proximally in the subclavicular region as a result of lead entrapment in the costoclavicular space by soft tissues or compression between the first rib and clavicle. Intravascular and tricuspid valve lead fracturing locations are very unusual. It is generally assumed that it arises from the crushing forces generated by high blood flow, valvular entrapment, and friction between leads.[10,11] The abrasive effect of micro-oganisms with biofilm formation on lead materials has also been suggested to explain lead fractures.[12] Direct bacterial damage may have contributed to lead fracturing in this case. Gradual decrease in pacing impedance weeks prior to the lead's fracture suggests insulation breach with wire exposition in our patient.

Conclusion

This case shows the relationship between colon cancer and streptococcal endocarditis. Both *Streptococcus* bacteraemia and streptococcal endocarditis should warrant the search for a gastrointestinal neoplasm.

This case also presents an association between endocarditis and lead fracture. It is widely accepted that mechanical forces are the main cause for intravascular lead fractures. Despite the absence of confirmatory post-mortem analyses in our case, the events preceding the lead rupture suggest that bacterial damage may have weakened the lead over time.

Long-term suppressive antibiotic therapy for CIED-related infections is associated with major complications and mortality as illustrated by the patient's fatal course caused by recurrent biliary sepses. Medical management alone should only be used in selected cases unsuitable for lead extraction (short life expectancy, very high operative risk, sepsis, etc.).[13] Complete hardware removal remains the

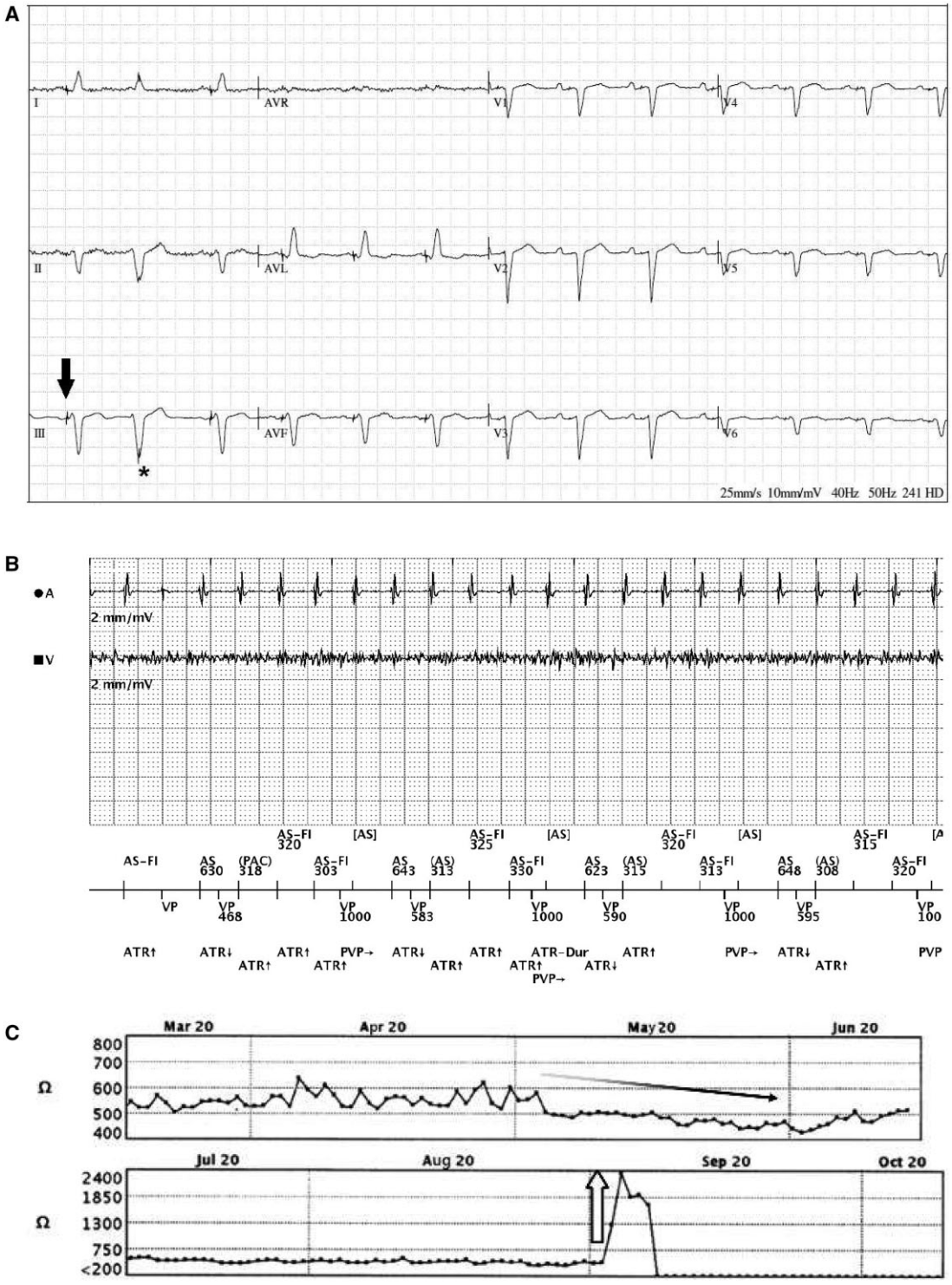

Figure 2 (*A*) Resting 12-lead electrocardiogram shows sinus rhythm with normal atrioventricular delay, loss of ventricular capture (arrow), ventricular undersensing (asterisk), and intrinsic ventricular depolarization. (*B*) Pacemaker electrogram shows atrial and ventricular electrogram with ventricular undersensing, lead noise, and loss of ventricular capture. (*C*) Pacemaker interrogation reveals a gradual decrease in ventricular pacing impedance in May 2020 (insulation breach) followed by a sudden increase in September 2020 (lead fracture). A, atrial lead; AS-FI, atrial sense in refractory period; ATR—Du ↑/↓, atrial tachycardia sense—duration started. ↑ count up/↓ count down; PAC, premature atrial contraction; PVP, postventricular atrial refractory period after a premature ventricular contraction; V, ventricular lead; VP, ventricular pace.

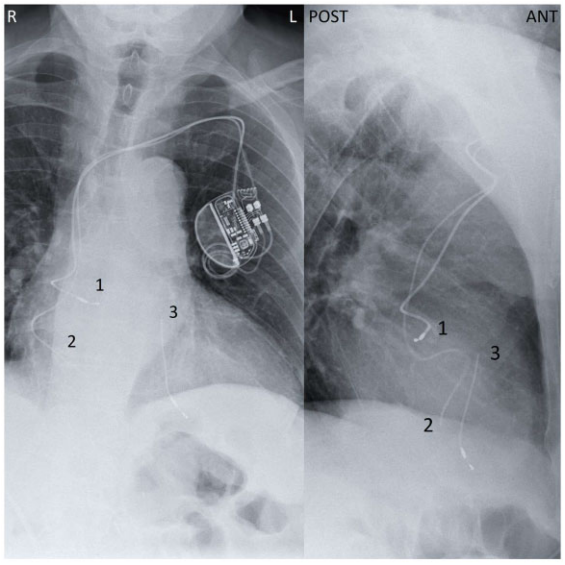

Figure 3 Chest X-ray locating the (1) atrial pacing lead, (2) proximal part of the ventricular lead, and (3) distal part of the ventricular lead. Dissociation of Parts 2 and 3 establishes lead rupture. *Left panel*: frontal view. *Right panel*: lateral view. (1) Atrial pacing lead. (2) Proximal part of the ventricular pacing lead. (3) Distal part of the ventricular pacing lead. ANT, anterior; L, left; R, right; POST, posterior.

requisite first-line therapy for patients with CIED-related infections, and should be referred to specialized centres.

Lead author biography

Dr Lorenzo Caratti di Lanzacco is a junior doctor pursuing a postgraduate in cardiology at the Catholic University of Louvain. He has special academic and clinical interests in Electrophysiology and Heart Failure medicine.

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 authors confirm that written consent for submission and publication of this case report including images and associated text has been obtained from the patient in line with COPE guidance.

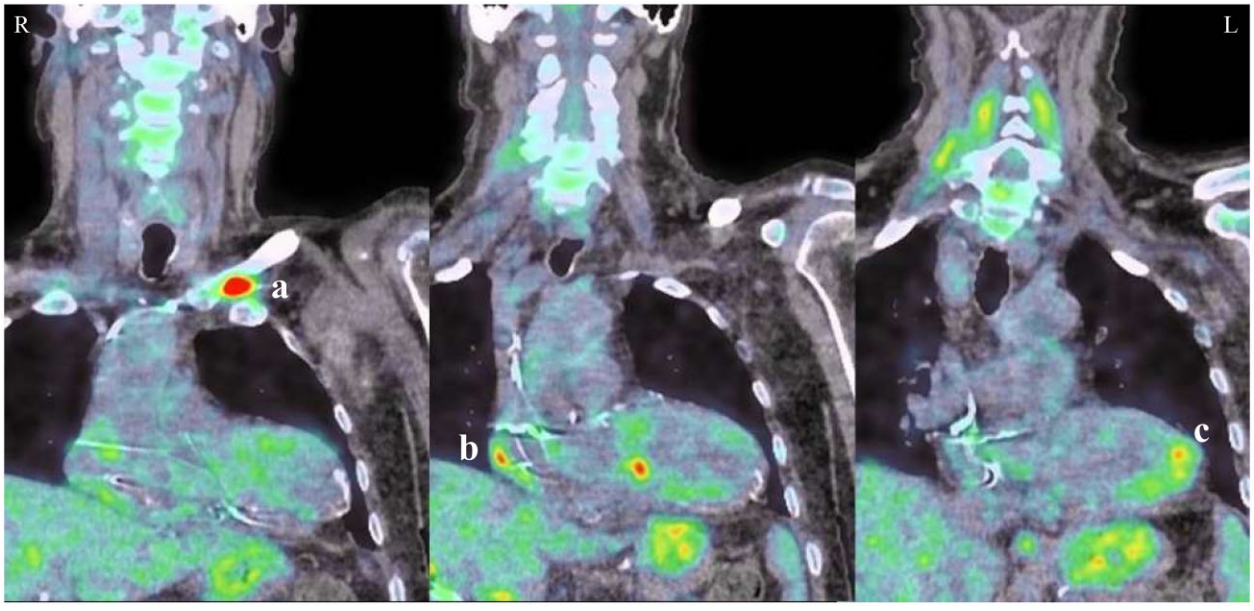

Figure 4 Positron emission tomography/computed tomography shows increased ^{18}fluorodesoxyglucose uptake on the extracardiac (*A*: box) and intracardiac (*B*: right atrium; *C*: right ventricle apex) portions of the pacemaker. These coronal images are compatible with pocket infection and lead endocarditis. L, left; R, right.

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