



Case Report

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Traps and Tricks About A 6-Years Old Cutibacterium Endocarditis

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ABSTRACT

Endocarditis of cardiac implanted electronic devices is a complex and serious disease. Commensal bacteria of the skin are commonly considered as contamination of blood culture. Herein, the authors report the case of a pacemaker endocarditis with cultures positive for *Cutibacterium acnes*. These were already found 3 and 6 years ago but not observed during hospitalisation. The PET-scan was of great help in guiding the diagnosis. The management was particularly difficult with 9 leads to extract. A sequential approach in the operating room from endovascular procedure to surgery under cardio-pulmonary bypass was used. A second procedure was necessary to extract a small forgotten foreign body already visible on the preoperative chest X-ray. This experience shows the importance of having a high level of suspicion in case of culture positive for *Cutibacterium* with intracardiac devices, and of looking for foreign bodies on the preoperative chest X-ray in case of a history of multiple rhythmological procedures.

Introduction

Endocarditis of Cardiac Implanted Electronic Devices (CIED) is becoming more common, with an incidence of 1.9 per 1000 device-years [1]. This is especially true when multiple CIEDs are present. These are serious diseases with a mortality rate of up to 20% [2]. The diagnosis and management of these diseases can be difficult. Older and multiple cardiac implanted devices are more prone to infection. As a result, cardiac implanted device endocarditis with these bacteria represents a particular nosological category with many pitfalls.

The authors report the case of a CIED endocarditis with cultures positive for *Cutibacterium acnes*, which was already found 3 and 6 years ago.

Cutibacterium acnes is a commensal bacterium found on the chest. Almost always due to contamination, it is rarely considered a patent infection [3]. It is responsible for only 0.3% of Infective Endocarditis (IE) but 2.5% of CIED endocarditis [4,5].

The authors report the case of a difficult diagnosis of CIED endocarditis. The management was particularly complex with 9 leads to remove. The authors used a sequential approach in the operating room, from

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endovascular procedure to surgery under cardiopulmonary bypass. A second procedure was necessary to extract a small foreign body, which was already visible on the preoperative chest X-ray.

Case Presentation

A 52-year-old woman was initially hospitalized due to a deterioration in general health and syncopes.

In her medical history, it is noteworthy that she had dilated cardiomyopathy and a heart rate that was difficult to balance by cardiologists with a pacemaker connected to three leads in the right atrium, the right ventricle, one in the coronary sinus, and two in the left pocket. The most recent procedure occurred a month ago: cardiac resynchronization with an implantable cardiac defibrillator on the right side, implanting leads in the right ventricle and atria. At the time, it was not possible to implant the leads in the coronary sinus due to technical difficulties.

She experienced chronic weight loss, asthenia, and frequent lipothymia. For one week, she had chills without fever. There were no local signs of inflammation. Blood tests revealed an inflammatory syndrome (CRP: 130 and neutrophils: 17). All three blood cultures showed only *Cutibacterium acnes*. Trans-thoracic and trans-oesophageal

echocardiography (TTE/TOE) revealed no signs of endocarditis. The authors concluded a high suspicion of CIED-endocarditis, although the criteria of the modified Duke's classification were not met. Upon reviewing the medical history, the authors discovered 4 and 2 blood cultures showing *Cutibacterium acnes* 6 years and 3 years ago, respectively. The PET-scanner showed two fixation sites: one on the long-known left subclavian artery, and the second one near the new pacemaker's casing on the right (Figure 1). The diagnosis of CIED-endocarditis with *Cutibacterium acnes* was made.

The heart team discussed this CIED-endocarditis and decided on the total extraction of the material with the implantation of a new epicardial 3-chamber pacemaker.

The first step of the surgery involved the extraction of the pacemaker placed on the right side. All the leads were then easily removed. On the left side, removal was not possible (even with the Cook system and the locking stylet). A median sternotomy was performed, during which the innominate trunk was opened between two lacks. All the proximal parts of the leads were withdrawn, but the distal extremities remained trapped in the right cavities. Subsequently, a cardiopulmonary bypass was conducted. Upon opening the right atrium, an evident CIED-endocarditis



Fig. 1. PET-scanner with two hot spots at late time.

The two arrows show two fixations, one on the left subclavian artery and one near the right pacemaker casing.

with multiple vegetations was encountered and removed.

Epicardial 3-chamber pacemakers with two additional defibrillator leads were installed.

The day after, a chest X-ray revealed two remaining lead parts near both the left and the right pocket. Fluoroscopic guidance assisted in completely

explanting them (Figure 2). Ultimately, a piece of a J-shaped probe and a lead's sheath were found. They were already visible on the preoperative chest X-ray (Figure 3).

After a 10-day hospital stay, the patient was discharged with antibiotics (Amoxicillin and Clindamycin; 14 days each). Six months later, the patient was doing well and showed no signs of infection.

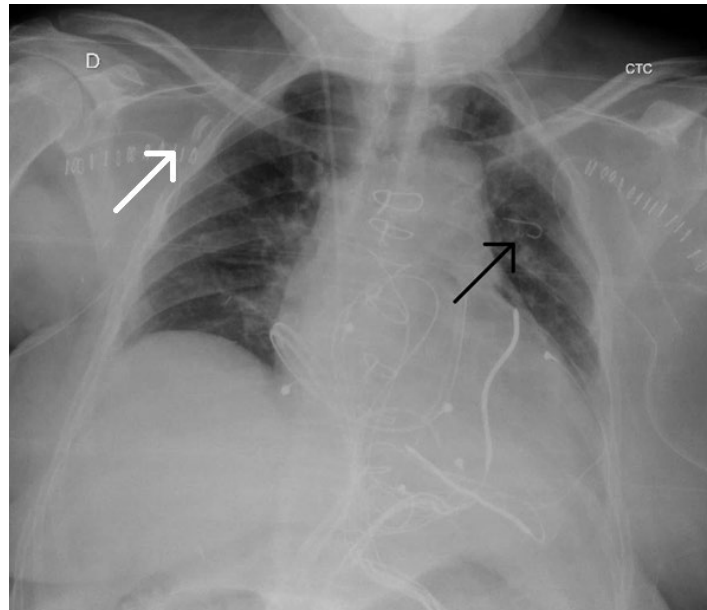


Fig. 2. Postoperative chest X-ray.

The arrow shows the foreign body remaining at the same place. It was the extremity of a probe. The white arrow shows lead's sheath.

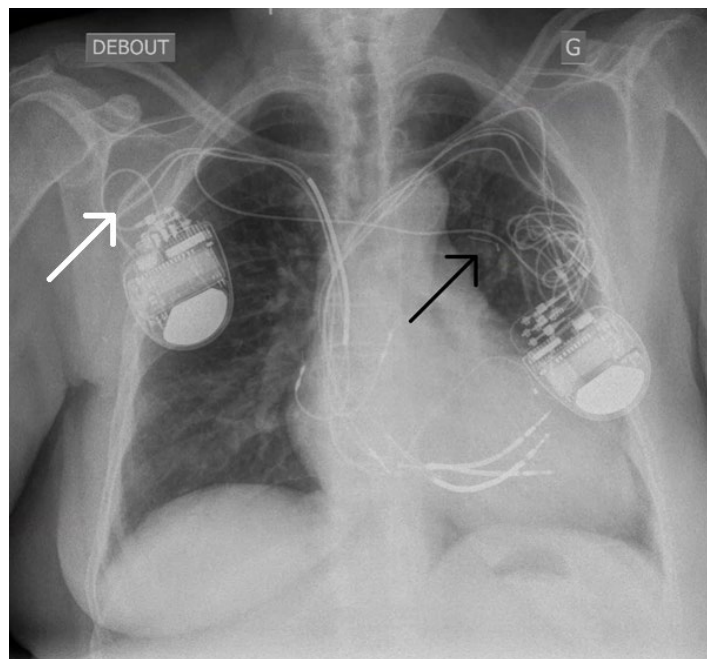


Fig. 3. Preoperative chest X-ray.

The black arrow shows a foreign body visible along the lead's path. The white arrow shows lead's sheath. In the presence of many tracks, strangulated bodies are more frequent but more difficult to see.

Discussion

Regarding the diagnostic

Cutibacterium acnes is a commensal anaerobic gram-positive bacillus typically found on the chest. It is almost always considered as blood culture contamination. Often, the results are not visible due to their very long culture time, given after discharge (up to 100 hours in this case). In a retrospective study, only 18 out of 522 blood cultures with *Cutibacterium acnes* were clinically significant bacteraemia. Among them, only one was an endocarditis [3]. It is responsible for only 0.3% of Infective Endocarditis (IE) and 2.5% of CIED-endocarditis [4,5]. However, *Cutibacterium acnes* endocarditis is almost always associated with intracardiac material (to the authors' knowledge, there is no study about the frequency of endocarditis in patients with positive *Cutibacterium acnes* blood culture, associated with intracardiac material).

A positive *Cutibacterium acnes* blood culture associated with intracardiac material prompts the authors to suspect CIED-endocarditis. These endocarditis have a late onset: from a few months to several years [6]. These properties led the authors to review blood cultures from previous hospitalizations.

Generally, the presentation is not specific, with a late diagnosis and severe intracardiac lesions [6]. Transoesophageal Echocardiography (TOE) has almost a 100% sensitivity and specificity to see vegetations [7]. This is less true in the case of CIED-endocarditis because of the anterior position of the right ventricle and the artefact caused by the leads. In this case, neither the Trans-Thoracic Echocardiography (TTE) nor the TOE showed any abnormalities.

To meet the modified Duke's criteria, the authors conducted a PET-scan. One study shows the advantage of a PET scanner in patients with an association of fever, negative endocarditis, and pacemaker: it allows the diagnosis of CIED-endocarditis in more than half of the patients [8]. The authors found two hotspots in the right and left subclavian arteries. There was no fixation inside the right cavities, which contradicted the operative findings. The PET-scan specificity is not sufficient for early infection [7]. In this case, it would have been difficult to conclude on the fixation on the new right leads. On the other hand, the old fixation on the left was more indicative of an established infection.

About the treatment

The decision was made to remove the leads in the

cardiac operative room. Indeed, the old, multiple [9], and ventricular [10] character was at high risk of failure by the simple endovascular approach. This allowed the authors to use a simple sequential approach with three extraction sites (from the least to the most invasive one).

The mortality rate for CIED endocarditis is around 20% [6]. Some teams report cases of *Cutibacterium acnes* endocarditis treated solely with antibiotics [11]. These were always patients with multiple comorbidities. Infectious recurrence is almost inevitable, with a high mortality rate [4]. Even though the patient had advanced dilated cardiomyopathy, it seemed that the benefit/risk ratio was in favor of total surgical extraction (with cardiopulmonary bypass).

The American Heart Association recommends the complete removal of the material: the pre- and post-operative analysis of the chest X-ray must be thoroughly done, especially when there is a large amount of material. Preoperatively, foreign bodies that are not part of the pacemaker need to be searched for, particularly along the leads (a small piece of probe in this case). This should avoid a second intervention. Even in the theater of open cardiac surgery, fluoroscopic control might prevent these kinds of complications.

The authors chose to place an epicardial pacemaker in this patient because of the surgery that allowed this approach, the numerous previous endovascular failures, and the immediate effectiveness of ventricular resynchronization. This was particularly the case when several leads had to be removed.

Conclusions

The authors report the case of a challenging management of a CIED endocarditis. There are several take-home messages for the reader.

Firstly, it is important to suspect an infection rather than just simple contamination when cultures positive for *Cutibacterium acnes* are encountered in patients with some CIED (and other commensal bacteria in general). Searching old blood culture results can greatly assist in these patients (even several years prior!). Transoesophageal Echocardiography (TOE) may not show present vegetation on cardiac leads. Therefore, the threshold to prescribe a PET-scan should be low in these situations.

Secondly, this case underscores the need to carefully examine the preoperative chest X-ray of patients who

have undergone multiple rhythmology procedures in the past to screen for any foreign bodies. Performing the procedure in the operative room allows for a change in plan if needed.

Ethical Considerations

Compliance with ethical guidelines

There were no ethical considerations to be considered in this article.

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Conflict of Interests

The authors have no conflict of interest to declare.

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References

- [1] Greenspon AJ, Patel JD, Lau E, et al. 16-year trends in the infection burden for pacemakers and implantable cardioverter-defibrillators in the United States 1993 to 2008. *J Am Coll Cardiol* 2011;58:1001-1006. <https://doi.org/10.1016/j.jacc.2011.04.033>
- [2] Baman TS, Gupta SK, Valle JA, Yamada E. Risk factors for mortality in patients with cardiac device-related infection. *Circ Arrhythm Electrophysiol* 2009;2:129-134. <https://doi.org/10.1161/CIRCEP.108.816868>
- [3] Park HJ, Na S, Park SY, et al. Clinical significance of Propionibacterium acnes recovered from blood cultures: analysis of 524 episodes. *J Clin Microbiol* 2011;49:1598-1601. <https://doi.org/10.1128/JCM.01842-10>
- [4] Lalani T, Person AK, Hedayati SS, et al. Propionibacterium endocarditis: a case series from the International Collaboration on Endocarditis Merged Database and Prospective Cohort Study. *Scand J Infect Dis*. 2007;39:840-848. <https://doi.org/10.1080/00365540701367793>
- [5] Bongiorno MG, Tascini C, Tagliaferri E, et al. Microbiology of cardiac implantable electronic device infections. *Europace* 2012;14:1334-1339. <https://doi.org/10.1093/europace/eus044>
- [6] Banzon JM, Rehm SJ, Gordon SM, Hussain ST, Pettersson GB, Shrestha NK. Propionibacterium acnes endocarditis: a case series. *Clin Microbiol Infect* 2017;23:396-399. <https://doi.org/10.1016/j.cmi.2016.12.026>
- [7] Habib G,ancellotti P, Antunes MJ, et al. 2015 ESC Guidelines for the management of infective endocarditis: The Task Force for the Management of Infective Endocarditis of the European Society of Cardiology (ESC). *Eur Heart J* 2015; 36:3075-3128. <https://doi.org/10.1093/eurheartj/ehv319>
- [8] Ploux S, Riviere A, Amraoui S, et al. Positron emission tomography in patients with suspected pacing system infections may play a critical role in difficult cases. *Heart Rhythm* 2011;8:1478-1481. <https://doi.org/10.1016/j.hrthm.2011.03.062>
- [9] Nof E, Epstein LM. Complications of cardiac implants: handling device infections. *Eur Heart J* 2013;34: 229-236. <https://doi.org/10.1093/eurheartj/ehs352>
- [10] Gula LJ, Krahn AD, Yee R, Skanes AC, Ghosh N, Klein GJ. Arrhythmia device lead extraction: factors that necessitate laser assistance. *Can J Cardiol* 2008;24:767-770. [https://doi.org/10.1016/S0828-282X\(08\)70681-0](https://doi.org/10.1016/S0828-282X(08)70681-0)
- [11] Tascini C, Bongiorno MG, Di Cori A, et al. Cardiovascular implantable electronic device endocarditis treated with daptomycin with or without transvenous removal. *Heart Lung* 2012;41:24-30. <https://doi.org/10.1016/j.hrtlng.2012.02.002>