

Staphylococcus lugdunensis infective endocarditis with perforation of the sinus of Valsalva: a case report

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Background

Staphylococcus lugdunensis endocarditis is a rare but fulminant disease.

Case summary

A 74-year-old female with a history of asymptomatic severe aortic valve stenosis and permanent atrial fibrillation presented with acute onset of fever (39.0°C). Electrocardiogram showed diffuse ST-segment elevation. She was hospitalized for further analysis. All blood cultures were positive for *Staphylococcus lugdunensis* and antibiotic treatment was started. Transthoracic echocardiography (TTE) showed known aortic valve stenosis without clear signs of endocarditis. The following day, a transoesophageal echocardiogram (TEE) showed a new moderate aortic valve regurgitation, new pericardial effusion (PE), and a thickened sinus of Valsalva (SOV) consistent with endocarditis with paravalvular involvement. Positron emission tomography-computed tomography was consistent with aortic valve endocarditis with paravalvular expansion. The patient was transferred to a tertiary referral centre for surgical treatment. On admission, patient was in shock and a second TTE revealed a new systolic and diastolic flow through the SOV to the right ventricle indicating SOV perforation. Additionally, there was flow in the PE suggestive of perforation of one of the cardiac chambers or large vessels. Emergent surgery showed extended infection with SOV perforation and a large perforation of the right ventricle. Ultimately, patient died during the operation because of extensive infection and refractory shock.

Conclusion

Staphylococcus lugdunensis endocarditis is a severe disease with poor response to conventional anti-microbial treatment, destructive complications requiring surgery, and has a high mortality risk.

Keywords

Infective endocarditis • *Staphylococcus lugdunensis* • Case report • Sinus of Valsalva perforation

ESC Curriculum

2.2 Echocardiography • 4.11 Endocarditis

Learning points

- *Staphylococcus lugdunensis* endocarditis has a fulminant clinical course and can lead to severe cardiac complications, such as a perforation of the sinus of Valsalva or perforation of the cardiac chambers, in several days despite adequate antibiotic therapy.
- *Staphylococcus lugdunensis* endocarditis should be treated with the same urgency and dedication as *Staphylococcus aureus* endocarditis.

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Introduction

Staphylococcus lugdunensis is a coagulase-negative *Staphylococcus* and was first recognized in 1988.¹

In the same year, a case of native valve endocarditis with this pathogen was reported which resembled the clinical course of *Staphylococcus aureus* endocarditis.² The formation of biofilm is thought to play a major role in the pathogenesis of *Staphylococcus lugdunensis* endocarditis since it allows attachment of the organism to host tissue and protects the organism from antibiotic therapy.³ In the last 30 years several case reports confirmed the fulminant behaviour of the disease which often results in acute and destructive infectious endocarditis.^{4–6}

Timeline

Timeline	Event
Day 0	<ul style="list-style-type: none"> ● Patient presented with a fever (39.0°C). Antibiotics were started and she was hospitalized.
Day 1	<ul style="list-style-type: none"> ● Blood cultures were positive for <i>Staphylococcus lugdunensis</i>. ● Transthoracic echocardiography (TTE) showed known aortic stenosis without clear signs of endocarditis.
Day 2	<ul style="list-style-type: none"> ● Transoesophageal echocardiography (TEE) showed new moderate aortic valve regurgitation, new pericardial effusion (PE), and a thickened sinus of Valsalva (SOV).
Day 3	<ul style="list-style-type: none"> ● Positron emission tomography-computed tomography (PET-CT) was consistent with aortic valve endocarditis with paravalvular expansion. ● Transfer to a tertiary hospital ● TTE showed signs of SOV perforation and flow in the PE. ● Emergency cardiothoracic surgery
Day 4	<ul style="list-style-type: none"> ● Patient died during the operation because of extended infection and refractory shock

Case presentation

A 74-year-old female presented with acute onset of fever (39°C). Her medical cardiac history was notable for asymptomatic severe aortic stenosis, hypertension, and atrial fibrillation. Her medication included dabigatran 150 mg twice daily, digoxin 0.0625 mg daily, and irbesartan 150 mg daily.

On presentation at the referring hospital (day 0), laboratory blood tests showed haemoglobin of 7.1 mmol/L, leukocytes of $19.0 \times 10^9/L$ creatinine of 60 $\mu\text{mol/L}$, glomerular filtration rate (GFR) 86 mL/min/1.73 m² and elevated levels of C-reactive protein at 160 mg/L (normal range, < 10 mg/L) and troponin-I of 1205 ng/L (normal range, < 14 ng/L). There were no endocarditis stigmata. Given her medical history of aortic valve stenosis, a known systolic murmur was present without a diastolic murmur. Electrocardiogram showed atrial fibrillation with diffuse ST-segment elevation indicative of pericarditis. TTE showed a preserved left ventricular ejection fraction of 55% and a known severe aortic valve stenosis with thickened leaflets. PE or vegetations was not observed. She was hospitalized at the cardiac care unit with an initial suspicion of peri-myocarditis.

The next day (day 1) all blood cultures were positive for *Staphylococcus lugdunensis* and the patient was prescribed

flucloxacillin 12 grams/day. No portal of entry was found. On day 2, a TEE showed a new moderate aortic valve regurgitation, new PE, and a thickened SOV consistent with endocarditis with paravalvular involvement.

A positron emission tomography-computed tomography (PET-CT) was performed to detect a possible portal of entry and potential additional peripheral embolic and metastatic infectious foci for adequate source control. The PET-CT was consistent with aortic valve endocarditis with paravalvular expansion towards the left ventricle (LV) base and showed increased pericardial and pleural effusion. There were no other metastatic infections.

During these first days patient developed shock with acute renal failure with creatinine levels reaching 337 $\mu\text{mol/L}$, a GFR of 12 mL/min/1.73 m² and she became anuric, for which noradrenaline was started.

The patient was transferred to a tertiary centre with surgery facilities. On arrival, she was in shock with a lactate of 5.3 mmol/L and an increasing need for a high noradrenaline dose. A second TTE on admission showed a heavily calcified, thickened aortic valve (Figures 1 and 2) without clear valvular vegetation. However, there was a mobile mass (± 12 mm) in the right ventricular outflow tract (RVOT) which was attached to the SOV consistent with vegetation. Furthermore, the colour Doppler showed a continuous flow through the SOV and to the RVOT at the site of the vegetation, suggestive of a perforation. Additionally, there was PE (maximum 15 mm) without echocardiographic signs of tamponade, but with a systolic flow in the pericardial space which suggests a perforation of one of the cardiac chambers or large vessels (see Supplementary material online, Videos S1 and S2).

Due to refractory cardiogenic shock and SOV perforation, the patient was sent for emergent cardiothoracic surgery. During first inspection, the pericardium was extensively infected and was easily ruptured by manipulation. The aortic valve was severely destructed and could be removed by just a forceps. The myocardium of the right ventricle was infected with a large central perforation of the free wall of the right ventricle. The aortic wall at the level of the annulus of the right coronary cusp was perforated and accompanied by abscess formation deep in the myocardial tissue. The perforation of the right ventricle was closed. Unfortunately, because of the severity of the infection which seemed to be beyond repair, with also refractory shock, the cardiothoracic surgeons had to stop the procedure. The patient died in the operating room.

Discussion

To the best of our knowledge, this is the first case of a *Staphylococcus lugdunensis* endocarditis with perforation of the SOV and right ventricle.

Staphylococcus lugdunensis endocarditis is a rare and fulminant disease. The exact incidence is unclear but current literature suggests that around 0.8–1.0% of infectious endocarditis is the consequence of *Staphylococcus lugdunensis* sepsis.⁵ Other coagulase-negative staphylococci, such as *Staphylococcus homidis* and *Staphylococcus epidermidis*, are known to have a non-destructive course.⁴ However, *Staphylococcus lugdunensis* endocarditis is different with a clinical course that closely resembles, or is even worse than *Staphylococcus aureus* endocarditis.⁶ If not properly detected and treated it can lead to significant morbidity and mortality, such as in this reported case. Previous studies suggest that surgical treatment is necessary in up to 70% of cases and mortality rates are up to 50%.^{5,6}

As noted in the presented case, extensive infection due to infective endocarditis may result in severe necrosis and rupture, and the subsequent development of rare aorto-cavitary perforation and fistulation. A previous study investigating 4681 episodes of infective endocarditis found that a total of 76 patients (1.6%) developed aorto-cavitary communications. These fistulae originate in similar rates from the aortic

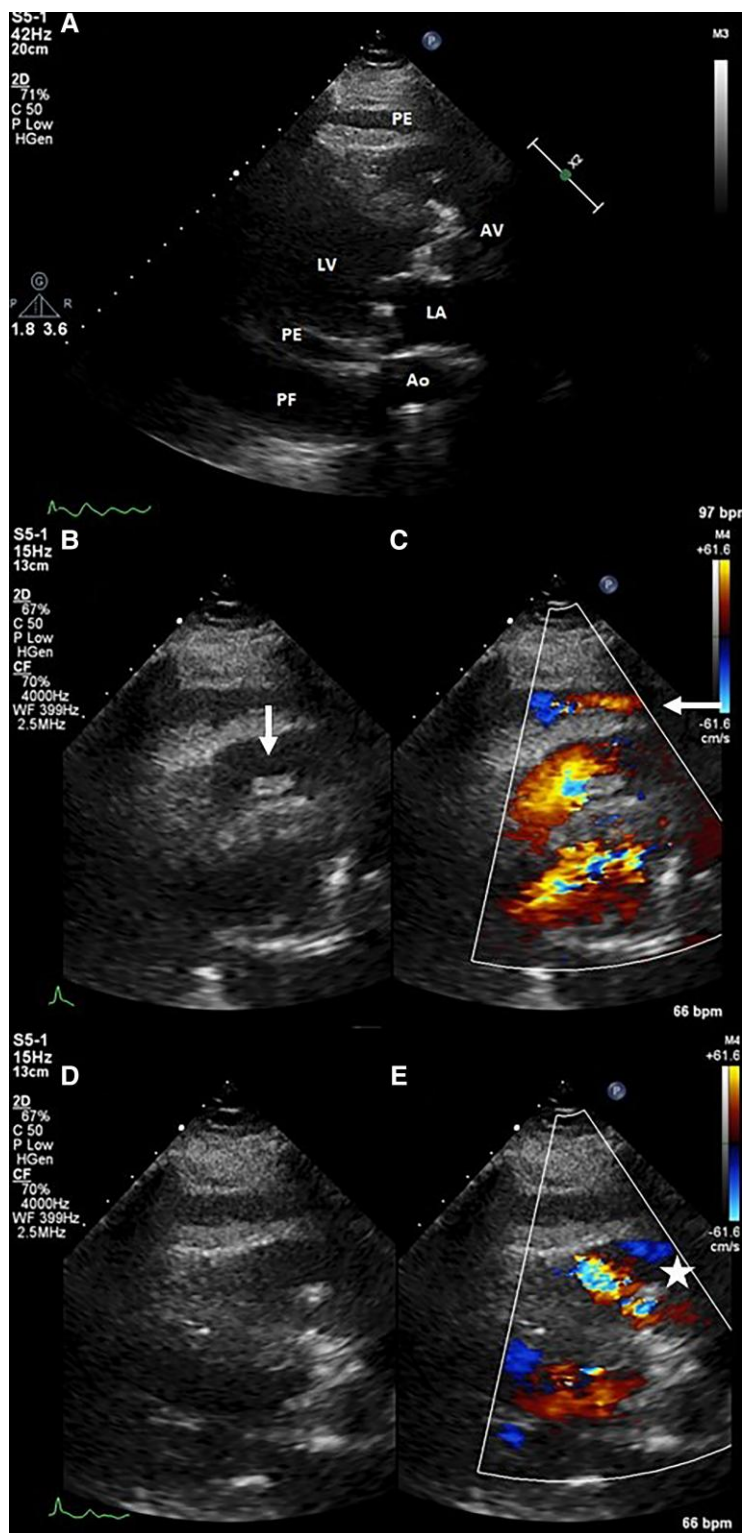


Figure 1 Parasternal long-axis view. (A) End diastolic image of the parasternal long-axis view of the aortic valve. A calcified and thickened aortic valve is seen. There is PE (maximum ± 12 mm) and pleural effusion. (B) Zoomed systolic image of the parasternal long-axis view of the aortic valve. A calcified aortic valve is seen with reduced opening consistent with aortic valve stenosis. Furthermore, there is a mobile mass (± 12 mm) in the right ventricular outflow tract (vertical white arrow) which is attached to the SOV consistent with vegetation. (C) Doppler echocardiography view of image B using ‘colour compare’ mode. There is a systolic flow of the PE (horizontal white arrow). (D) Diastolic image of the parasternal long-axis view of the aortic valve. (E) Doppler echocardiography view of image D using ‘colour compare’ mode. There is diastolic flow through the SOV to the right ventricular outflow tract consistent with an SOV perforation (asterisk). Abbreviations: Ao, descending aorta; AV, aortic valve; LA, left atrium; LV, left ventricle; PE, pericardial effusion; PF, pleural fluid.

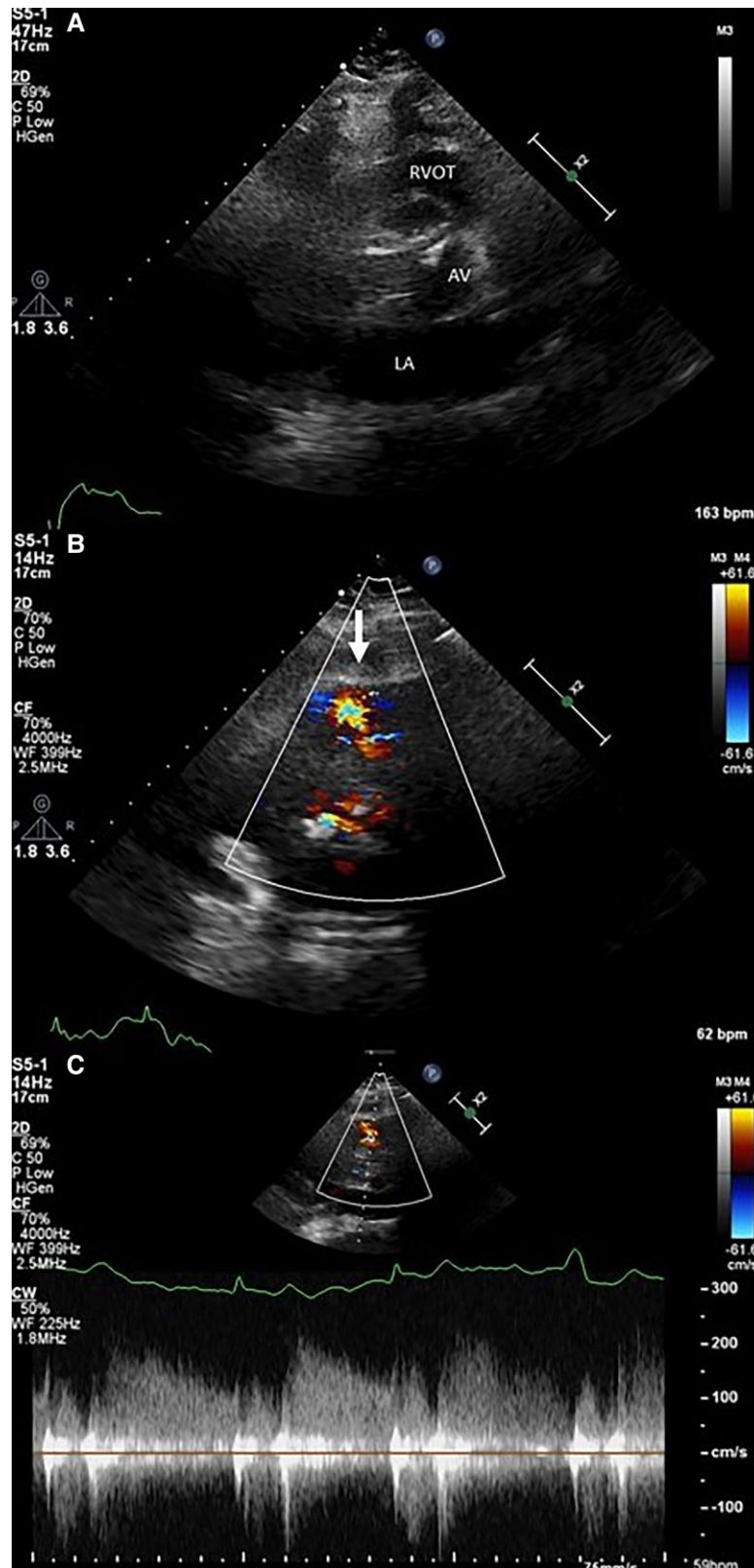


Figure 2 Parasternal short axis view. (A) Systolic image of the parasternal short axis view of the aortic valve. A calcified tricuspid aortic valve is seen. (B) Parasternal short axis view with Doppler echocardiography. There is a systolic flow in the RVOT indicative of SOV perforation. (C) Continuous wave Doppler of the flow in the SOV. There is a systolic and diastolic flow with a maximum velocity of 2 m/s. Abbreviations: AV, aortic valve; LA, left atrium; RVOT, right ventricular outflow tract.

sinuses and the four cardiac chambers are equally involved as well.⁷ Occurrence of aorto-cavitary communications results in further clinical deterioration and haemodynamic instability, requiring surgery in up to 87% of these patients.⁷

Flow in the pericardium, assessed by echocardiography, is suggestive of an abnormal communication between the pericardial space and one of the cardiac chambers or large vessels.⁸ In this case the intrapericardial flow was probably due to a large perforation of the right ventricular myocardium seen during surgery. Furthermore, PE in infective endocarditis is associated with more severe infections and worse prognosis and therefore always warrants further evaluation.^{9,10}

In order to improve patient care we recommend frequent echocardiographic evaluation in all patients with *Staphylococcus lugdunensis* endocarditis since the clinical course can be detrimental in a very short time. Furthermore, we advise that all patients at diagnosis should be transferred to a tertiary referral centre with surgical treatment options so that early surgical treatment can be initiated in case of an operation indication.

In conclusion, the current case of *Staphylococcus lugdunensis* endocarditis highlights the severity of the disease and the importance of early surgical treatment since severe cardiac complications can develop rapidly.

Lead author biography



Dr. Max J.M. Silvis studied medicine at the Radboud University of Nijmegen (Nijmegen, The Netherlands). After obtaining his medical degree, he started a PhD trajectory at the Cardiology department of the University Medical Center of Utrecht (Utrecht, the Netherlands). He defended his thesis entitled ‘Coronary Artery Disease: Inflammatory Pathways and Interventions’ in 2022. He continues his medical and scientific career in the field of Cardiology as he started his cardiology training at the Radboud University

Medical Center in October 2022.

Supplementary material

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

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

Consent: In accordance with COPE guidelines, the widower of the patient involved signed informed consent including consent to publish the anonymized data.

Conflicts of interest: All authors declare that there is no conflict of interest.

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Data availability

The data underlying this article will be shared on reasonable request to the corresponding author.

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