

CASE REPORT

***Staphylococcus lugdunensis* bacteraemia secondary to an infected intracardiac fragment of a vascular access device**

Lisa Micallef Grimaud¹, Josephine Bigeni², Matthew Mercieca Balbi², Nina Nestorova³, Walter Busuttill⁴,
Tonio Piscopo¹

¹ Infectious Diseases Unit, Mater Dei Hospital, Malta

² Department of Medicine, Mater Dei Hospital, Malta

³ Department of Microbiology, Mater Dei Hospital, Malta

⁴ Cardiac Surgical Unit, Mater Dei Hospital, Malta

ABSTRACT

We report a case of *Staphylococcus lugdunensis* bacteraemia following infection of a fragmented totally implantable vascular access device. This report highlights the virulence of *S. lugdunensis* especially in immunosuppressed patients with prosthetic devices as well as the importance of distinguishing between contamination and blood stream infection when this organism is cultured from blood. The importance of prompt removal of implantable vascular access devices when they are no longer required, to prevent catheter related infections is also reflected in this report. *J Microbiol Infect Dis* 2015;5(2): 85-88

Key words: *Staphylococcus lugdunensis*, bacteraemia, vascular access devices.

Damar yolu açma aparatının intrakardiyak parçasında gelişen enfeksiyona sekonder *Staphylococcus lugdunensis* bakteriyemisi

ÖZET

Parçalara ayrılmış implante edilebilir damar yolu açma aparatı enfeksiyonunu takip eden bir *Staphylococcus lugdunensis* bakteriyemisi olgusunu bildirdik. Bu olguda özellikle protez cihaz kullanan immünitese baskılanmış hastalarda gelişen *S. lugdunensis*'in virülansı vurgulamakla birlikte bu mikroorganizmanın kandan izole edildiğinde kontaminasyon/enfeksiyonunu ayırımının önemine dikkat çekildi. İhtiyaç kalmadığında damar yolu açma aparatlarının hemen çıkarılmasının kateter enfeksiyonlarının önlenmesinde arz ettiği önem ayrıca belirtildi.

Anahtar kelimeler: *Staphylococcus lugdunensis*, bakteriyemi, damar yolu açma aparatı

INTRODUCTION

Whilst most Coagulase negative *Staphylococci* (CoNS) are regarded as harmless contaminants, the importance of *Staphylococcus lugdunensis* as a virulent pathogen cannot be undermined.¹⁻⁴ We report a case of *S. lugdunensis* bacteremia following infection of a fragmented totally implantable vascular access device (TIVAD) in a patient with rheumatoid arthritis. Despite several reports of *S. lugdunensis* endocarditis in the literature, to our knowledge *S. lugdunensis* infecting a fragmented TIVAD has not been described.

CASE REPORT

Our patient is a 60 year old woman who presented with a six week history of fatigue, fever and night sweats. This patient had an extensive medical history. At the age of 27 she underwent a total hysterectomy and oophorectomy for ovarian cysts. In subsequent years, she developed bowel adhesions causing intestinal obstruction which necessitated repeated small bowel resections. One such operation which she had undergone 12 years prior to this presentation, led to a prolonged period of hospitalization during which the lady required a TIVAD due

Correspondence: Lisa Micallef Grimaud, 81 La Cascara, Triq il-Qasam, Swieqi, Malta
Email: lisamicg@gmail.com

Received: 01 June 2014, Accepted: 08 October 2014

Copyright © Journal of Microbiology and Infectious Diseases 2015, All rights reserved

to difficult venous access. This lady also had a history of thromboembolic events including pulmonary embolism and a cerebrovascular accident. She also suffered from rheumatoid arthritis which had been controlled on methotrexate. This was stopped by her general practitioner at the onset of fever.

On admission to hospital, the patient had a fever of 38.6°C. Her pulse rate was 110 beats per minute whilst blood pressure was 126/70 mmHg. She had herpes simplex labialis and the reservoir of the TIVAD was noted in the right infra-clavicular region. There was no erythema at the site. Oxygen saturation was 96% on atmospheric air, and chest and cardiovascular examinations were normal. Her abdomen revealed multiple surgical scars and there was no organomegaly. Rheumatoid nodules were palpable at the elbows but she had no active synovitis.

Her blood investigations revealed a white cell count of $14.8 \times 10^9/L$ and raised inflammatory markers (ESR: 65mm, CRP 170 mg/L). Chest roentgenography (Fig.1) showed detachment of the TIVAD catheter from the reservoir, the tip of the catheter being in the region of the right atrium. The electrocardiogram was normal. Three sets of blood cultures were taken after which empirical treatment with levofloxacin was commenced.

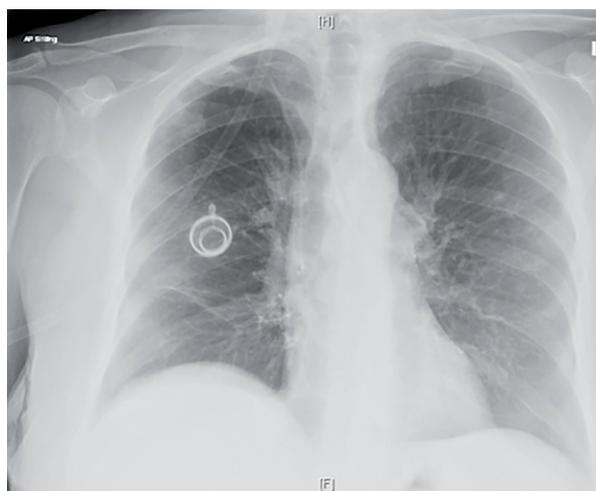


Fig.1. Chest X-Ray of the patient

All three sets of blood cultures flagged positive using the BacT/ALERT (bioMerieux) blood culture system. Gram stain and microscopy were performed on a smear prepared using a loopful of broth from each bottle, confirming the presence of Gram-positive cocci. Two blood agar plates and a

MacConkey agar plate were inoculated with a loopful of the broth. The blood agar plates were respectively incubated, aerobically and anaerobically, in a CO₂-fortified atmosphere, while the MacConkey agar plate was incubated aerobically without CO₂ enrichment. Colony growth was evaluated using Gram staining which again confirmed the presence of Gram-positive cocci. The catalase test was positive; coagulase and DNAase tests resulted negative. These findings were indicative of a coagulase-negative Staphylococcus. The latter was detected in the first set of blood culture bottles, but since it was presumed to be a contaminant further identification tests were not carried out. When however, two subsequent sets of blood cultures yet again detected the same bacterium, identification and sensitivity testing was carried out using the VITEK 2 system using GP and AST P580 (bioMerieux) cards, respectively. The growth of *Staphylococcus lugdunensis* was confirmed which was sensitive to the penicillins, fluoroquinolones, macrolides, aminoglycosides, tetracyclines, glycopeptides as well as rifampicin, fusidic acid, clindamycin and tigecycline.

Meanwhile, a transthoracic echo confirmed the presence of a vegetation in the right atrium. This was better visualised on a transoesophageal echo which revealed a highly mobile vegetation measuring 1cm at its longest axis attached to the tip of the fragmented line. An incidental patent foramen ovale was also noted.

In view of the above findings, levofloxacin was switched to vancomycin, gentamicin and rifampicin. Therapeutic levels of vancomycin and gentamicin were regularly monitored.

A CT of the chest was performed in order to accurately localize the tip of the catheter. This showed cavitation containing gas in the subcutaneous tissue of the right upper chest wall in place of the proximal aspect of the previously inserted line. The line was identified with its proximal end in the right atrium, extending to the superior vena cava and right brachiocephalic vein.

In the first instance, the reservoir of the TIVAD was removed uneventfully. Fragments of the reservoir were vortexed in 10 mls Ringer's solution. One hundred microlitres (100 µl) of the solution were then pipetted and streaked using a spreading technique onto a blood agar plate and incubated overnight in 5% CO₂-enriched aerobic conditions at 35-37°C. The growth was deemed to be significant since >10 CFU (colony forming units) were detected. Gram stain, microscopy and biochemical identification

tests on the resultant colonies were repeated and once again, using the VITEK 2 system, the growth of *Staphylococcus lugdunensis* was confirmed.

Preparations were then made for the detached line to be removed. The large size of the vegetation and the fact that it was very mobile, as well as the patient's patent foramen ovale, made retrieval by percutaneous sling method too risky and it was thus decided to approach the fragmented catheter by open surgery. The preoperative X-ray showed that the line had migrated further to the pulmonary artery. In fact, at surgery, the catheter was palpable through the pulmonary artery. The patient was placed on cardiopulmonary bypass, an incision made in the pulmonary artery and the line extracted using forceps (Fig.2).



Fig. 2. Extracted catheter with vegetation

The patient received intravenous antimicrobial therapy consisting of vancomycin, gentamicin and rifampicin for 28 days. Rifampicin was then switched to the oral formulation and levofloxacin added for a further 2 weeks. The patient made a good recovery and remains well.

DISCUSSION

First described by Freney in 1988, *Staphylococcus lugdunensis* was originally regarded as a harmless skin commensal and contaminant similar to most of the coagulase negative staphylococci (CoNS).⁵ However, its role as an important human pathogen later emerged as it was found to be responsible for invasive infections with life threatening complications. Hence *Staphylococcus lugdunensis* has often been referred to "a wolf in sheep's clothing".¹⁻⁴

This virulent CoNS can cause aggressive destruction behaving more like *Staphylococcus aureus* and it has been associated with a wide array of infections including bacteremia, skin and soft tissue infections, endocarditis, osteomyelitis, prosthetic joint infections, peritonitis, endometritis, endophthalmitis and abscesses.²

Staphylococcus lugdunensis has a propensity to infect prosthetic material, and review of the literature also suggests that there is a link between immunosuppression and infection with this organism.^{2,3} In the case described above, our patient had been on methotrexate for treatment of her rheumatoid arthritis. She also had a TIVAD, a device which is generally associated with low rates of infection and other complications.⁶ In our case, however, fragmentation of the TIVAD occurred at some point in time prior to admission. The patient did not clearly describe an event which might have been associated with rupture. However, many years had passed since it had been inserted, and long term use together with material fatigue and degradation are regarded to be predisposing factors for rupture of TIVADs.⁷

This case report highlights the importance of distinguishing between contamination and blood stream infection (BSI) once the growth of a CoNS is detected. Early identification of a BSI is important since this leads to prompt initiation of antimicrobial treatment, reduction in the rate of complications, decreased hospital stay, and most importantly decreased mortality. On the other hand, identifying contamination is important to avoid unnecessary use of antimicrobials and the emergence of antimicrobial resistance. However, distinguishing blood culture contaminants from true bacteraemias caused by CoNS is not straightforward.^{8,9}

Whilst growth of a CoNS from more than one blood culture bottle is indicative of a BSI, studies have revealed that up to one third of patients with a true BSI have only one positive blood culture. In one study, the presence of Systemic Inflammatory Response Syndrome (SIRS) criteria (temperature $>38^{\circ}\text{C}$ or $<36^{\circ}\text{C}$; heart rate $>90/\text{min}$; respiratory rate $>20/\text{min}$; leucocytosis or leucopenia) and the presence of an implant or device made the presence of a blood stream infection more likely. In this same study, it was suggested that the presence of 3 SIRS criteria or 2 SIRS criteria and a central venous catheter, makes the presence of a BSI more likely when there is a single blood culture positive for CoNS;⁸ In another large study carried out in Finland, the pres-

ence of a central venous catheter also made a BSI more likely when CoNS was cultured from blood. However, this study maintained that clinical and laboratory parameters may not always provide help in determining the significance of CoNS blood culture findings.⁹

In the case described above, the fact that the patient had a vascular access device as well as her being febrile, tachycardic and having a leucocytosis, made a blood stream infection very likely from the outset. The clinical details accompanying our patient's first set of blood cultures could not be traced unfortunately, but insufficient clinical details could have led to the microbiologists not pursuing any further identification tests once CoNS was detected from the first set of blood cultures. Thus, the importance of communicating the essential clinical details to the microbiology laboratory cannot be overemphasized. This report also highlights the importance of identifying CoNS down to species level. The fact that CoNS, identified as *Staphylococcus lugdunensis*, was cultured from two further blood culture sets as well as from the reservoir of the Tl-VAD, made the diagnosis of a BSI indisputable in the case described.

CONCLUSION

Staphylococcus lugdunensis is frequently isolated in the bacteriology laboratory but it should not be considered a contaminant a priori. This report emphasizes the importance of proper interpretation of such a culture within the context of the clinical picture. The importance of maintaining a high index of suspicion, especially when prosthetic material is involved and/or one is dealing with immunosuppressed patients, is crucial so as not to misinter-

pret growth of *Staphylococcus lugdunensis* as presumed contamination.

Furthermore, the removal of implantable vascular access devices without delay when they are no longer required is central to prevent the deleterious complications of catheter related infections and mechanical problems such as fragmentation.

REFERENCES

1. Choi S, Chung J, Lee EJ, et al. Incidence, Characteristics, and outcomes of *Staphylococcus lugdunensis* bacteraemia. *J Clin Microb* 2010;48:3346-3349.
2. Babu E, Oropello J. *Staphylococcus lugdunensis*: The coagulase negative staphylococcus you don't want to ignore. Expert review of anti-infective therapy. 2011;9:901-907.
3. Frank KL, del Pozo J, Patel R. From clinical microbiology to infection pathogenesis: How daring to be different works for *Staphylococcus lugdunensis*. *Clin Microbiology Review* 2008;21:111-33.
4. Herchline T, Ayers L. Occurrence of *Staphylococcus lugdunensis* in consecutive clinical cultures and relationship of isolation to infection. *J Clin Microbiology* 1991;29:419-421.
5. Freney J, Brun Y, Bes M, et al. *Staphylococcus lugdunensis* sp.nov. and *Staphylococcus schleiferi* sp.nov., Two Species from Human Clinical Specimens. *Int J System Evolution Microbiol* 1988;38:168-172.
6. Royle T, Davies R, Gannon M. Totally implantable vascular access device-20 years' experience of implantation in cystic fibrosis patients. *Ann R Coll Surg Engl* 2008;90:679-684.
7. Di Carlo I, Biffi R. Totally Implantable Venous Access Devices: Management in Mid- and Long-term Clinical Setting. Italia: Springer-Verlag, 2012:201-204.
8. Elzi L, Babouee B, Vogeli N, et al. How to discriminate contamination from bloodstream infection due to coagulase-negative staphylococci: a prospective study with 654 patients. *Clin Microbiol Infect* 2012;18:E355-E361.
9. Rahkonen M, Luttinen M, Koskela. True bacteremias caused by coagulase negative *Staphylococcus* are difficult to distinguish from blood culture contaminants. *Eur J Clin Microbiol Infect Dis* 2012;31:2659-2644.