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Case study

Infective endocarditis caused by *Pseudomonas stutzeri* in a patient with Marfan syndrome: Case report and brief literature review

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ABSTRACT

Invasive infections due to *Pseudomonas stutzeri* have rarely been described and mainly occur in immunocompromised individuals. We report a case of infective endocarditis caused by *P. stutzeri* after previous cardiac surgery in a Lebanese patient with Marfan syndrome. We review the literature and conclude that this pathogen may be of particular medical relevance in the Mediterranean Basin.

Introduction

Human infections due to non-fermentative bacteria of the genus *Pseudomonas* account for considerable morbidity and mortality worldwide. While most infections are caused by *P. aeruginosa*, other *Pseudomonas* spp. may also give rise to severe infections, albeit less frequently. *P. stutzeri* has been reported as a pathogen that may occasionally cause severe deep-seated infections such as vertebral osteomyelitis and meningitis in immunocompromised individuals [1].

Here, we describe a case of infective endocarditis caused by *P. stutzeri* in a Lebanese patient with Marfan syndrome, and we review the literature to elucidate specific factors related to severe *P. stutzeri* infections.

Case report

A 22-year-old male patient with Marfan syndrome was referred to our hospital in Germany for severe and symptomatic aortic insufficiency. He had undergone valve-preserving aortic root replacement (valve reimplantation) and concomitant mitral repair with implantation of an annuloplasty ring three years earlier in his home country, Lebanon. After having lived in Germany for two years, he had developed severe and symptomatic aortic insufficiency. Echocardiography on admission showed severe aortic insufficiency and dilatation of the left ventricle (end-diastolic diameter 80 mm). In addition, floating structures were found on echocardiography that were consistent with vegetations on the mitral annuloplasty ring (Fig. 1). Diagnostic work-up for possible endocarditis was performed.

At the time of presentation, the patient was afebrile, there was mild elevation of leukocytes and a C-reactive protein (CRP) of 25 mg/L (normal value: < 5 mg/L). Blood cultures were drawn, and empiric antimicrobial treatment with intravenous ampicillin/sulbactam was initiated. Assuming a device-related endocarditis, the patient underwent repetition of surgery. After dissection of adhesions and connection to cardiopulmonary bypass, the heart was explored. The aortic valve showed signs of cusp retraction and low commissural height, there was no evidence of active endocarditis. Repair was considered not feasible, and the valve replaced with a 25 mm mechanical prosthesis (St. Jude Medical). The mitral valve was inspected; the valve itself was found intact, but there were gross vegetations located on the annuloplasty ring. The ring was excised; no new annular stabilization was performed. The postoperative course was unremarkable.

After surgery, vancomycin was added to the empiric antimicrobial regimen to cover pathogens giving rise to prosthetic device infections, in particular coagulase-negative staphylococci. Microbiologic cultures of the surgically removed annuloplasty ring grew *P. stutzeri*. This was confirmed by polymerase chain reaction (PCR) assays, and no additional pathogens were identified.

Thus, the diagnosis of definite infective endocarditis was made [2]. The antimicrobial treatment was immediately adjusted to an anti-pseudomonal regimen consisting of piperacillin/tazobactam and gentamicin. The isolated *P. stutzeri* strain was found susceptible to several antimicrobials including piperacillin/tazobactam, ceftazidime, and the carbapenems. Minimal inhibitory concentrations (MICs) were lower for ceftazidime than for piperacillin/tazobactam, and treatment was thus re-adjusted to ceftazidime intravenously for four weeks. Gentamicin

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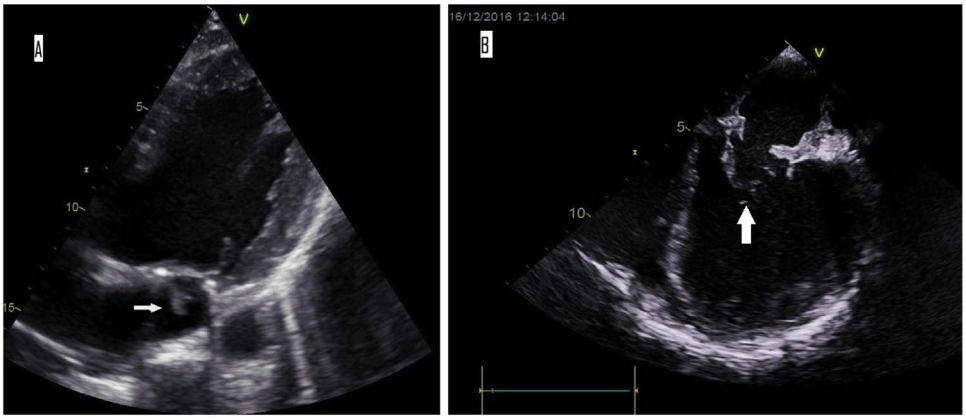

Fig. 1. (A) Transthoracic echocardiography of a patient with *P. stutzeri* endocarditis, apical view of the left heart demonstrating a floating structure on the mitral ring, which is suggestive of a bacterial vegetation (arrow). (B) Transesophageal echocardiography of the same patient: long section of the mitral valve, demonstrating the same floating structure (arrow).

Table 1
Overview about the published cases of human *P. stutzeri* infections in the international literature (1995–2017), providing clinical information on the site of infection and the country where the infection occurred. Note: Cases published until 1994 have previously been summarised in another review [9].

Clinical description of *P. stutzeri* infection	No. of reported cases	Year of publication	Country	Reference
Bacteremia in a patient with acute leukemia	1	1995	Spain	[16]
Meningitis in a patient with HIV infection	1	1996	Spain	[17]
Pleural empyema in a cirrhotic patient	1	1996	Spain	[18]
Bacteremia related to a bullous skin eruption	1	1996	Australia	[19]
Community-acquired pneumonia with empyema	1	1997	Spain	[20]
Endophthalmitis after cataract surgery	1	1998	Czech Republic	[21]
Community-acquired vertebral osteomyelitis	1	1999	USA	[22]
Prosthetic joint infection of the knee in a patient with acute promyelocytic leukemia	1	2000	Israel	[23]
Panophthalmitis with orbital abscess	1	2001	USA	[24]
Endocarditis caused by *P. stutzeri* and *Streptococcus salivarius*	1	2002	Spain	[13]
Bacteremia in a patient with ecthyma gangraenosum and systemic vasculitis	1	2004	France	[25]
Community-acquired pneumonia with empyema in a young boy	1	2004	Turkey	[26]
Bacteremia	4	2004	Taiwan	[27]
Endophthalmitis after cataract surgery	1	2006	India	[28]
Pneumonia in an HIV patient	1	2006	United Kingdom	[10]
Posttraumatic osteitis of the left femur	1	2006	Morocco	[29]
Brain abscess in a child after previous neurosurgery	1	2006	USA	[30]
Neonatal sepsis in a baby born to a mother with pre-eclampsia and prematurely ruptured membranes	1	2007	Saudi Arabia	[31]
Posttraumatic knee infection in a child	1	2007	Israel	[32]
Bacteremia cases in a teaching hospital over a 1-year period, all except one in patients with chronic diseases (COPD, renal insufficiency)	8	2007	Brazil	[15]
Conjunctivitis after cataract surgery	1	2008	India	[33]
Relapse of *P. stutzeri* endocarditis four years after initial presentation	1	2009	France	[14]
Fatal community-acquired meningitis in an immunocompetent 73-year old patient	1	2009	Turkey	[34]
Two cases of ventilator-associated pneumonia and one case of diabetic ulcer infection	3	2009	Italy	[35]
Peritonitis	1	2010	Turkey	[36]
28 patients with detection of *P. stutzeri* from a sterile site (n = 18, blood; n = 4, peritoneal fluid; n = 3, conjunctiva; n = 2, synovial fluid; n = 1, cerebrospinal fluid)	28	2012	Israel	[37]
Peritonitis in a 82-year old patient on continuous ambulatory peritoneal dialysis (CAPD)	1	2013	South Korea	[38]
Necrotizing pneumonia in a patient with concomitant pulmonary tuberculosis	1	2014	Taiwan	[11]
Corneal ulcer in a 25-year old patient after previous neurosurgery	1	2015	India	[39]
Bacteremia with carbapenem-resistant *P. stutzeri* in a patient undergoing hematopoietic stem cell transplantation	1	2015	China	[40]
Meningitis during vedolizumab treatment in a patient with Crohn's disease	1	2015	USA	[41]
Prosthetic vascular graft infection and prosthetic osteomyelitis of the tibia	2	2016	Canada	[42]
Prosthetic valve endocarditis (present case)	1	2017	Lebanon	–

was stopped after 7 days.

Follow-up echocardiographic studies were unremarkable. All blood cultures taken during the further in-hospital stay of the patient remained negative. The patient was discharged home in good general condition on oral ciprofloxacin for additional four weeks. Upon several follow-up outpatient visits after completion of the antimicrobial treatment, the patient was asymptomatic and no irregularities were seen on echocardiography.

Discussion

The case reported here is the fourth description of an endocarditis due to the gram-negative bacterium *P. stutzeri*, and the first one in a patient with Marfan syndrome. Gram-negative pathogens account for less than 10% of all cases of infective endocarditis worldwide [3,4], and endocarditis due to *Pseudomonas* spp. is a rarity. However, previous reports indicated that unusual bacterial pathogens such as *Abiotrophia defectiva* [5], *Brucella suis* [6] and *Neisseria mucosa* [7] might be of particular relevance as agents of infective endocarditis in patients with Marfan syndrome.

P. stutzeri was first described by Burri and Stutzer in 1895 [8]. This gram-negative, rod-shaped, aerobic, and oxidase-positive bacterium is naturally found in soil and water [1]. When a clinical specimen grows the organism, it is commonly considered as colonization or

contamination [9]. True human infections with this bacterium occur particularly in immunocompromised patients with chronic comorbidities or a history of previous surgery, and are frequently related to prosthetic devices. Indeed, it has also been hypothesized that *P. stutzeri* infections may develop more easily in inflamed tissue, as it was observed in two patients being treated for pulmonary tuberculosis with considerable parenchymal destruction, who developed complicating superinfections with *P. stutzeri* [10,11].

A review published in 1994 concluded that *P. stutzeri* rarely causes clinical disease and generally responds well to targeted antimicrobial treatment [9]. Based on the detection of this pathogen in our case, we performed a literature review of all clinical cases of infections due to *P. stutzeri* being published in MEDLINE-indexed journals between 1 January 1995 and 31 May 2017. The major findings are summarized in Table 1. We noted a considerable increase in cases of *P. stutzeri* infections. Whereas only 20 patients had been reported in the literature until 1994, our case represents the 73rd description of an infection due to this pathogen in the 23-year period ever since. The most common types of infection were bacteremia (n = 35, 48%), ocular infections (n = 8, 11%), pneumonia and pleural empyema (n = 7, 10%) as well as bone and joint infections (n = 7, 10%). Peritonitis and infections of the central nervous system were also detected. However, only three cases of *P. stutzeri* endocarditis have been reported previously [12–14], two of which were related to prosthetic valves and occurred up to 4 years after cardiac surgery. In our case, the source of the patient's *P. stutzeri* infection could not be unambiguously identified, but there might have been a relation to the patient's previous surgery in Lebanon.

We found a striking geographical distribution of *P. stutzeri* infections, with 62% of all globally reported cases being detected in the Mediterranean Basin, most frequently in Israel (n = 30), Spain (n = 5), Italy (n = 3) and Turkey (n = 3). Of note, very few cases were reported from the United States of America and Western Europe. One series from Brazil reported eight cases of bacteremia in a single hospital, which might have been related to a common source of contamination [15]. The three previous cases of endocarditis had been reported from France, Israel and Spain, all of which are countries in the Mediterranean Basin. Our patient was from Lebanon, which further underscores the potential geographical pattern of *P. stutzeri* infection. It remains unclear whether these observations relate to higher rates of contaminated medical equipment and devices in this region or whether there are specific biological factors favouring the growth of *P. stutzeri* in such environments. Further investigations are warranted to elucidate these associations.

We conclude that *P. stutzeri*, even though frequently considered as contaminant, can give rise to potentially severe infections, including endocarditis. A literature review suggests that *P. stutzeri* might occur more frequently in Israel and other countries of the Mediterranean Basin.

Conflicts of interest

All authors report no conflict of interest relevant to this article.

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