IDCases 17 (2019) e00562

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcr

Case report

Stenotrophomonas maltophilia right-sided endocarditis in an adult patient with uncorrected congenital heart disease

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ARTICLE INFO

Article history:
Received 4 April 2019
Received in revised form 10 May 2019
Accepted 10 May 2019

Introduction

Stenotrophomonas maltophilia is a Gram-negative, aerobic, glucose non-fermenting, motile bacillus.

The pathogen was first isolated from pleural effusion and named Bacterium booker in 1943. After then it was classified into different groups and, in 1993, reclassified in group *Stenotrophomonas* [1]. *Stenotrophomonas maltophilia* can form a biofilm, an ability to attach and grow on the surface by extracellular matrix formation; the higher the level of the biofilm, the more resistant to most antibacterials [2]. World Health Organization list this organism as one of the leading multiple-drug-resistant organisms (MDRO) for hospital situations (website). However, this ability to be resistant to the first line of antimicrobials does not correlate with the prior use of antibacterials and is completely related to intrinsic factors, such as beta-lactamase production [3]. *Stenotrophomonas maltophilia* is regarded as a low virulence, opportunistic and significant nosocomial pathogen that which requires background factors for infection, such as immunodeficiency states, intravenous indwelling catheters or any situation that breaks the mucosa and skin barriers [4].

Infection with *Stenotrophomonas maltophilia* is nosocomial in 76% of cases and community acquired in the remaining [5]. It most commonly associated with respiratory infections especially in patients with underlying lung diseases such as cystic fibrosis and/or chronic obstructive pulmonary disease. While *Stenotrophomonas maltophilia* might infect other organs, endocarditis is quite rare. Bacteremia with this pathogen are accompanied with quite high mortality. In one retrospective study, bacteremia with *Stenotrophomonas maltophilia* were compared with *Pseudomonas aeruginosa* and *Acinetobacter* species in regard to a secondary 30-day all-cause mortality rate, which were found to be 33.1%, 21.5% and 17.3% respectively [6]. Therefore, early diagnosis, as well as quick and appropriate antibacterial therapy, can be crucial and lifesaving. Combined with conventional blood culture, the polymerase chain reaction (PCR) could be helpful for an earlier diagnosis. Hence, prescription of antimicrobial of choice should be justified, as soon as possible, that is still trimethoprim-sulfamethoxazole, as monotherapy or as a part of combination therapy [7].

Case report

A 26 year old man, known case of surgically uncorrected ACHD; small size membranous type ventricular septal defect (VSD) with pulmonary-systemic shunt ratio less than 1.5:1.0, presented with one-month history of fever, profuse sweating, lassitude and marked weight loss. His problem started when he was abroad and despite outpatient follow up, his illness became worse over time. He had received some analgesics and a short course of unknown oral antibacterial. Continuation of fever, pain in the right upper abdomen and severe pleuritic pain in the right upper chest for seven days were reported. On arrival to our ward, when he came back home, he appeared to be toxic, acutely ill, pale, with shallow and rapid respiration because of pleuritic pain. On physical examination, high grade fever with engorged neck veins, tender hepatomegaly, palpable spleen, splinter haemorrhage and palatal petechiae were found, but no clubbing, was detected. Also, there was palpable thrill over the precordium in addition to an audible high-grade pansystolic murmur in the left lower sternal border. Sinus tachycardia in electrocardiography and haziness in the upper part of right lung on chest X-Ray were noted. Leukocytosis and moderate anaemia were reported. Sedimentation rate, c -reactive protein (CRP) and rheumatoid factor, were elevated (78 mm/h, 24 mg/L and 28 IU/ml respectively). The transthoracic echocardiography (TTE) as well as

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https://doi.org/10.1016/j.idcr.2019.e00562

transoesophageal echocardiography (TEE) showed severe tricuspid regurgitation due to destroyed & flailed tricuspid valve of all three leaflets with many attached echodensities in several sizes, some small and some more than two centimetres. Also, there was a small size echodensity attached at the right side of VSD (Fig. 1).

After blood cultures were obtained, empirical intravenous (IV) triple antibacterials (ampicillin, vancomycin and amikacin) was started. On the same day, he underwent emergency cardiac surgery and the tricuspid valve (TV) was replaced with a bioprosthetic valve in addition to VSD patch closure (American Heart Association/ American College of Cardiology (AHA/ACC) Class IIa; Level of Evidence C). Three days later, two separate blood culture results confirmed *Stenotrophomonas maltophilia*. Based on an antibacterial sensitivity panel result, the therapy had switched to IV trimethoprim-sulfamethoxazole and oral tetracycline plus rifampin. He became afebrile on the fifth post-operative day. In the interim, TTE revealed a large amount of pericardial effusion impending to cardiac tamponade in addition to a large size echogenic mass, with possibility of clot, behind the right atrial wall. His chest re-opened and the pericardial drainage of bloody effusion and clot removal was performed uneventfully. The patient survived with combination of emergency surgery and four weeks of antibacterial therapy which helped the patient leave the hospital uncomplicatedly, despite the 33% mortality and 80% morbidity rate of endocarditis with *Stenotrophomonas maltophilia* that have been reported in the literature [8]. Within six months' post discharge follow up, he returned to the same weight before his illness and there was neither clinical nor laboratory evidence of PVE. Monthly echocardiography indicated well-functioning and vegetation-free bioprosthetic tricuspid valve.

Discussion

Infective endocarditis (IE) is an uncommon infectious disease with one-year mortality rate of 25% [9]. Despite the use of Prevention of Infective Endocarditis Guidelines 2007, from the (AHA) / (ACC), the incidence of IE has increased from 2007 to 2011 and reached to 15 per 100,000 in the United States [10]. Since the worldwide outbreak of rheumatic fever has declined [11], other heart diseases that have a longer lifespan with medical and surgical advances, have a higher percentage among endocarditis-susceptible groups. Patients with

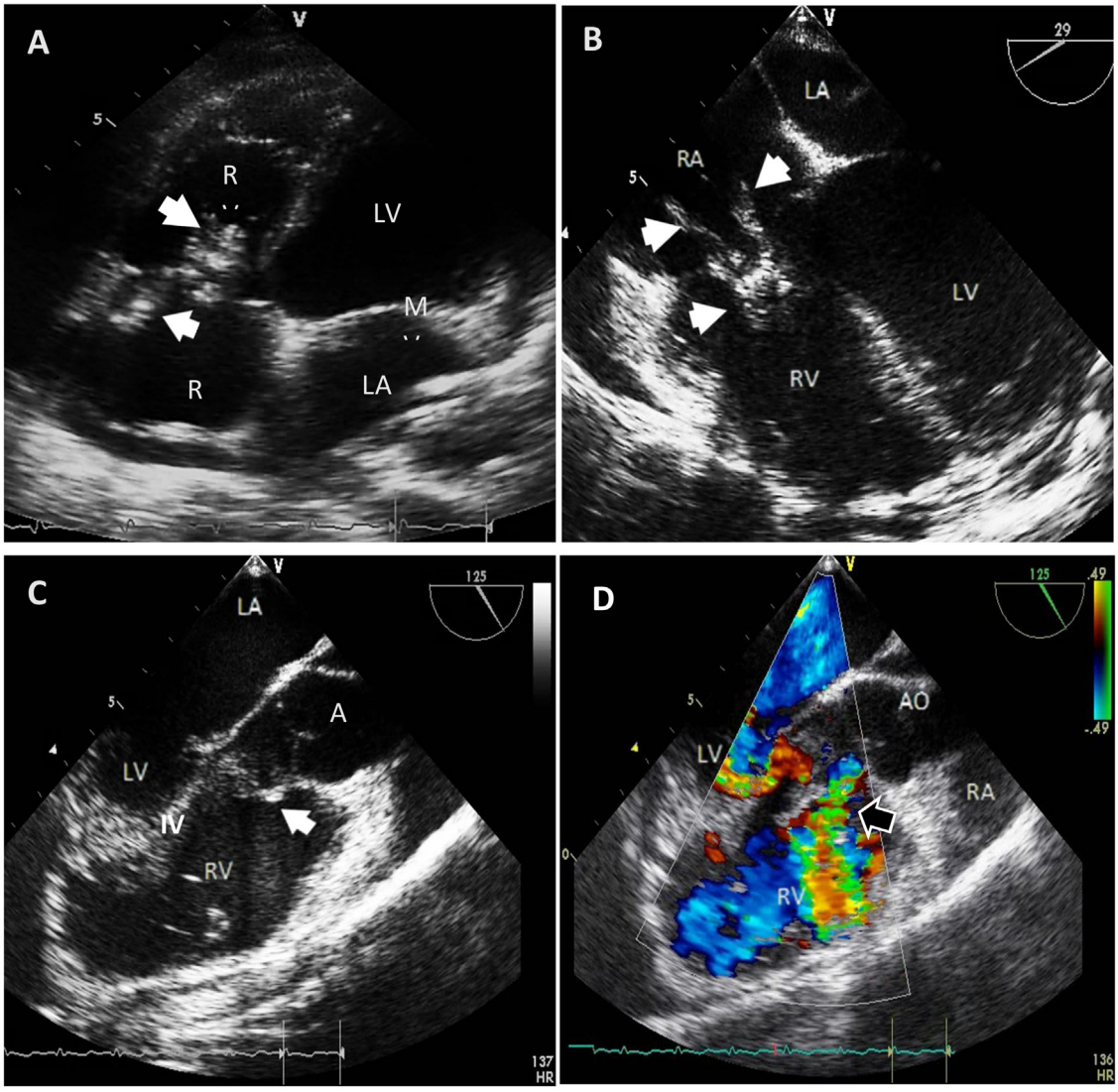

Fig. 1. Images A & B images demonstrate destroyed & flailing tricuspid valve (TV), in addition to attached & mobile vegetations in different sizes (white arrow heads). Image C demonstrates a small size vegetation attached to the right side of VSD (black arrow head with white margins). **A**. Transthoracic (TTE) apical four chamber off-axis view. **B**. Transeophageal (TEE) four chamber view (mid esophageal. **C**. TTE four chamber view (modified mid esophageal). **D.** Color Doppler TEE (in the same view as shown in image C). **Abbreviations**: AO: aorta. IVS: interventricular septum, LA: left atrium, LV: left ventricle, MV: mitral valve, RA: right atrium, RV: right ventricle.

surgically corrected adult congenital heart disease (ACHD), namely repaired with prosthetic material, palliative shunt or conduit, are in the top list of IE-sensitive group.

The incidence of endocarditis in patients with ACHD is remarkable: between 0.9 and 1.3 cases/1,000 patient-years, as opposed to the non-CHD population: 5–7/100,000 patient-year [12]. Among the ACHD patients, the most common underlying condition susceptible to endocarditis is VSD with an incidence of 1.9 cases/1000 patient years with predominance of right sided endocarditis. Bacterial endocarditis in ACHD patients has been reported with almost any bacterium. The most frequently implicated causative organism are streptococcus (37%) [13]. However, IE due to *Stenotrophomonas maltophilia* is scarce, with less than fifty reported cases worldwide to date, which caused up to 33% mortality among patients [14]. Three out of forty-five patients reviewed and reported by Subhani Sh. et al, had ACHD, while all of them underwent corrected cardiac surgery before, as opposed to our patient. Here, we present a known case of uncorrected ACHD presented with acute S. maltophilia endocarditis who was properly managed and left the hospital with no sequela. To the authors' knowledge, this is the first case report of infective endocarditis caused by *Stenotrophomonas maltophilia* in a patient suffering from uncorrected congenital heart disease.

Conflict of interest statement

The authors have indicated they have no financial relationships relevant to this article to disclose.

We would like to declare the contribution of each authors as follows. Sharifkazemi MB: patient's cardiologist, literature reviewer and write of the manuscript, Nemati MH, patinet's cardiac surgeon and critically reviewed the manuscript, Moarref AL. fellowship in Echocardiograhy who did the first TTE & TEE, also reviewed the manuscript

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

World Health Organization. Antimicrobial resistance: global report on surveillance. WHO, 2014. http://apps.who.int/iris/bitstream/10665/112642/1/9789241564748.

Acknowledgments

The authors would like to thank Sharifkazemi MM, from LSE in UK, for critically edit the article.

Recommended Reviewers:

References

[1] Al-Anazi KA, Al-Jasser AM. Infections caused by Stenotrophomonas maltophilia in recipients of hematopoietic stem cell transplantation. Front Oncol 2014;25(August (4)):232.
[2] Brooke JS. New strategies against Stenotrophomonas maltophilia: a serious worldwide intrinsically drug-resistant opportunistic pathogen. Expert Rev Anti Infect Ther 2014;12(January (1)):1–4.
[3] Chang YT, Lin CY, Chen YH, Hsueh PR. Update on infections caused by Stenotrophomonas maltophilia with particular attention to resistance mechanisms and therapeutic options. Front Microbiol 2015;02(September), doi:http://dx.doi.org/10.3389/fmicb.2015.00893.
[4] Araoka H, Baba M, Yoneyama A. Risk factors for mortality among patients with Stenotrophomonas maltophilia bacteremia in Tokyo, Japan, 1996-2009. Eur J Clin Microbiol Infect Dis 2010;29(May (5)):605–8.
[5] Lai CH, Chi CY, Chen HP, Chen TL, Lai CJ, Fung CP, et al. Clinical characteristics and prognostic factors of patients with Stenotrophomonas maltophilia bacteremia. J Microbiol Immunol Infect 2004;37:350–8.
[6] Hotta G, Matsumura Y, Kato K, Nakano S, Yunoki T, Yamamoto M, et al. Risk factors and outcomes of Stenotrophomonas maltophilia bacteraemia: a comparison with bacteraemia caused by Pseudomonas aeruginosa and Acinetobacter species. PLoS One 2014;9(11)e112208, doi:http://dx.doi.org/10.1371/journal.pone.0112208 Published online 2014 Nov 6.
[7] Kempf VA, Mandle T, Schumacher U, Schäfer A, Autenrieth IB. Rapid detection and identification of pathogens in blood cultures by fluorescence in site hybridization and flow cytometry. Int J Med Microbiol 2005;295(1):47–55, doi:http://dx.doi.org/10.1016/j.ijmm.2004.12.006.
[8] Subhani S, Patnaik AN, Barik R, Nemani L. Infective endocarditis caused by Stenotrophomonas maltophilia: a report of two cases and review of literature. Indian Heart J 2016;68(September (Suppl. 2)):S267–70.
[9] Ambrosioni J, Hernandez-Meneses M, Téllez A, Pericàs J, Falces C, Tolosana JM, et al. The changing epidemiology of infective endocarditis in the twenty-first century. Curr Infect Dis Rep 2017;19(5):21.
[10] Pant S, Patel NJ, Deshmukh A, Golwala H, Patel N, Badheka A, et al. Trends in infective endocarditis incidence, microbiology, and valve replacement in the United States from 2000 to 2011. J Am Coll Cardiol 2015;65(19):2070.
[11] Seckeler MD, Hoke TR. The worldwide epidemiology of acute rheumatic fever and rheumatic heart disease. Clin Epidemiol 2011;22(February (3)):67–84.
[12] Montanaro C, Dimopoulos K, Shore DF. Infective endocarditis in patients with congenital heart disease: when, where and how. Int J Cardiol 2017;15(December (249)):171–2.
[13] Moore B, Cao J, Kotchetkova I, Celermajer DS. Incidence, predictors and outcomes of infective endocarditis in a contemporary adult congenital heart disease population. Int J Cardiol 2017;15(December (249)):161–5.
[14] Satish M, Mumtaz MA, Bittner MJ, Valenta C. Stenotrophomonas maltophilia endocarditis of an implantable cardioverter defibrillator lead. Cureus J Med Sci 2019;11(3)e4165, doi:http://dx.doi.org/10.7759/cureus.4165.