Case report

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

Mohammadbagher Sharifkazemi\(^a,b\), Mohammadhassan Nemati\(^b\), Alireza Moarref\(^a\)

\(^a\) Department of Cardiology, Shiraz University of Medical Sciences, Postal code: 719371331, Iran

\(^b\) Department of Surgery, Shiraz University of Medical Sciences, Iran

**A R T I C L E   I N F O**

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**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 mono-therapy or as a part of combination therapy [7].

**Case report**

A 26 year old man, known case of surgically corrected 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
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 afibrile 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/1000 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 Echocardiography 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.

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