Available online at www.sciencedirect.com

ScienceDirect

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

Case Reports

Infective endocarditis caused by *Stenotrophomonas maltophilia*: A report of two cases and review of literature

Shaik Subhani [a,*], *Amar N. Patnaik* [b], *Ramachandra Barik* [b], *Lalita Nemani* [b]

[a] *Senior Resident, Department of Cardiology, Nizam's Institute of Medical Sciences, Hyderabad 500082, India*
[b] *Department of Cardiology, Nizam's Institute of Medical Sciences, Hyderabad 500082, India*

ARTICLE INFO

Article history:
Received 28 December 2013
Accepted 30 July 2015
Available online 14 January 2016

Keywords:
S. maltophilia
Infective endocarditis
Nosocomial
In-hospital habitat

ABSTRACT

Stenotrophomonas maltophilia is known for nosocomial habitat. Infective endocarditis due to this organism is rare and challenging because of resistance to multiple broad-spectrum antibiotic regimens. Early detection and appropriate antibiotic based on culture sensitivity reports are the key to its management. We report the diagnosis, treatment, and outcome of two cases of infective endocarditis caused by *S. maltophilia*.

© 2015 Cardiological Society of India. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Background

Stenotrophomonas maltophilia (*S. maltophilia*) is a nonfermentative, gram-negative, aerobic bacillus that is widely distributed in the nature. In a hospital setting, it has been found in water-related sources and contaminated medical equipments.[1–3] Although *S. maltophilia* is not highly virulent, its treatment is challenging because of its resistance to multiple antibiotics. Therefore, the relentless progression of patient's underlying illness adds to higher causalities.[4–7] Infective endocarditis due to *S. maltophilia* is very rare. Only 41 cases have been reported so far worldwide, most of which required surgical treatment. In this report, we share our experience of two cases of infective endocarditis managed by culture-sensitive antibiotics.

2. Case report

2.1. *Case 1*

A 35-year-old man was admitted to our hospital 3 months back with history of fever with chills for five days. The patient had undergone PBMV for severe rheumatic mitral stenosis 2 weeks prior to this episode. No other predisposition was found. There was no history of dental procedures or injections of intravenous drugs. On physical examination, blood pressure was 110/70 mmHg, pulse rate 85/min, and body temperature of 38 °C. Cardiac examination revealed loud S1 and P2 with grade III mid-diastolic murmur at the apex. The remaining physical examination was unremarkable. There were no peripheral signs of

* *Corresponding author.*
E-mail address: drsubhani03@gmail.com (S. Subhani).
http://dx.doi.org/10.1016/j.ihj.2015.07.048
0019-4832/© 2015 Cardiological Society of India. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

infective endocarditis. Laboratory tests showed that his Hemoglobin was 12.8 gm/dl, white blood cell (WBC) was 18,900/L, erythrocyte sedimentation rate (ESR) was 52 mm in first hour, and urinalysis was normal. Renal parameters were normal. Cardiomegaly was apparent in chest radiography.[12] Electrocardiogram revealed a normal sinus rhythm. The transthoracic echo demonstrated moderate mitral stenosis, severe eccentric mitral regurgitation, which was not there previously, and suspicious vegetation was present on mitral valve. The transesophagial echocardiogram revealed freely mobile sessile vegetation of size 5 mm × 5 mm over both anterior and posterior mitral leaflets (Fig. 1). The patient was started with a regimen of ceftriaxone and gentamycin. *S. maltophilia* was identified on blood culture and the antibiotics were changed to co-trimoxazole and levofloxacin as per culture sensitivity report. After 1 week of antibiotics, patient became afebrile, and repeat transesophagial echo after 2 weeks revealed disappearance of vegetation (Fig. 2). Antibiotics were continued for 6 weeks. He was discharged successfully.

2.2. Case 2

A 40-year-old man was admitted for fever and chill for a period of 2 months following mechanical valve replacement (27 mm Carbo-Medics) of mitral valve for severe rheumatic mitral stenosis. No other comorbidities were found. There was no history of dental procedures or injections of intravenous drugs. At presentation, he was drowsy, pale, and edematous with raised jugular venous pulsation. Blood pressure was 90/60 mmHg, pulse rate 120/min, and body temperature was 38 °C. Cardiac examination revealed soft S1, normal S2 with metallic heart sound. There was no audible murmur. Chest examination revealed bilateral basal crepitations. Glasgow Coma Scale (GCS) at presentation was E3 M5 V4 with no focal neurological deficits. The rest of the physical examination was unremarkable. Laboratory tests showed that Hb was 7.6 gm/dl, WBC 17,500/L, and ESR was 80 mm/hr. The renal parameters were elevated with urea of 65 mg/dl and serum creatinine of 3.0 mg/dl. Arterial Blood Gas (ABG) revealed metabolic acidosis. Increased cardiothoracic ratio and features suggestive of pulmonary edema were observed on chest radiography. Electrocardiogram revealed sinus tachycardia. The transthoracic echo demonstrated large mobile vegetation of 2 cm × 1.6 cm in size on prosthetic mitral valve (Fig. 3). There was mild paravalvular leak with partial dehiscence, moderate mitral regurgitation, moderate tricuspid valve regurgitation, moderate pulmonary arterial hypertension, and moderate left ventricular dysfunction. The patient was empirically started on vancomycin and ceftriaxone. Initially, blood cultures grew MRSA sensitive to only teicoplanin, gentamycin, and linezolid. Fever continued with spikes. Supportive treatment was given in the form of inotropic support, vasodilators, and peritoneal dialysis for rapidly worsening renal dysfunction. By the time, the repeat blood culture could identify the culprit bacteria to be *S. maltophilia*, the patient died of septicemia and renal dysfunction.

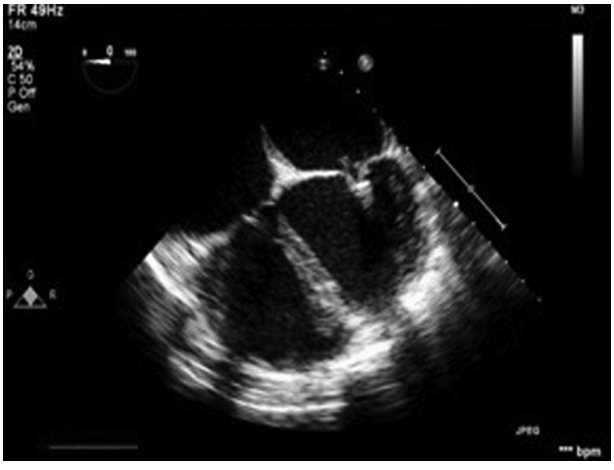

Fig. 1 – Transesophagial echocardiography (TEE) image is showing two mobile vegetations over anterior and posterior mitral leaflet before treatment of case number one.

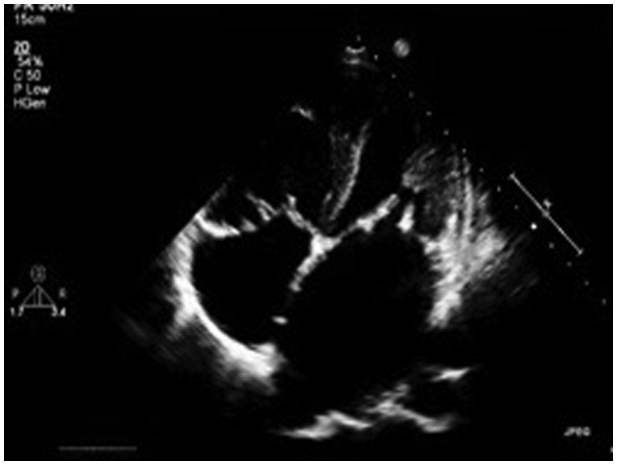

Fig. 2 – Transesophagial echocardiography (TEE) image is showing disappearance of vegetation after 2 weeks of culture sensitive-based antibiotic regimen of case one.

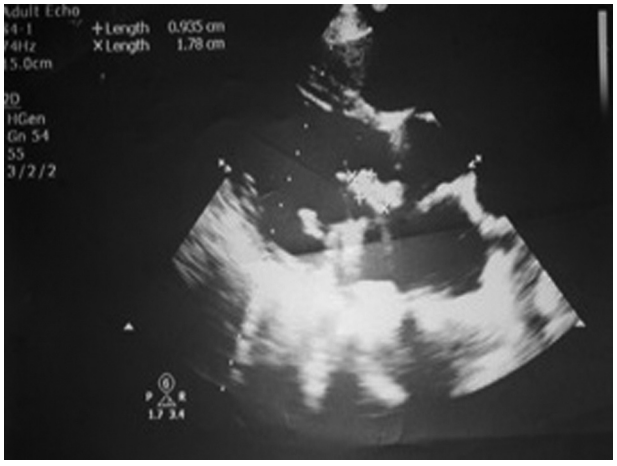

Fig. 3 – Transesophagial echocardiography (TEE) image is showing mobile vegetation over prosthetic mitral leaflet of case 2 who died in hospital.

Table 1 – The brief summary of worldwide experience of *Stenotrophomonas maltophilia* infectious endocarditis.

Case	Ref.	Clinical profile			Management		Complications	Outcomes
		Age/Sex	Predisposing factors	Valve involved	MM	Surgery		
1	[9]	26/M	Recent valve replacement (<1 month)	PMV, ASD patch	CHL, KAN, COL	Yes	Multiple septic emboli	D
2	[9]	30/M	Recent valve replacement (<1 month)	PMV, ASD patch	CHL	No	None	C
3	[9]	65/F	Cystoscopy, valve replacement (7 months)	PMV	CAR, GEN, KAN, CHL, PEN, POL	Yes	Persistent bacteremia	C
4	[9]	35/M	Recent valve replacement (early), rheumatic carditis	PMV	CAR, GEN, SXT	No	None	C
5	[9]	38/M	None	PMV	STR, PEN	No	Septic emboli, MI	D
6	[9]	22/M	IVDU	PAV	CAR, AMK, SXT	Yes	CHF, septic emboli	C
7	[9]	31/F	IVDU, dental treatment	PAV	CAR, KAN, SXT	Yes	Perivalvular abscess	C
8	[9]	57/M	IVDU, rheumatic carditis	NAV, NMV	POL, SXT	No	Septic emboli	C
9	[9]	25/M	None	VSD repair	GEN, CHL	No	NR	D
10	[9]	33/M	IVDU, aortic stenosis, atrial fistula	NAV, NTV	TIC, MOX, SXT	Yes	CHF, myocardial abscess	D
11	[9]	NR	CVC	NTV	NR	NR	NR	D
12	[9]	NR	NR	NAV	NR	NR	NR	D
13	[9]	NR	CVC	NAV	NR	NR	NR	C
14	[9]	33/M	IVDU, dental treatment	PAV	SXT, AMC, GEN	Yes	Perivalvular abscess	C
15	[9]	56/M	Recent valve replacement (early)	PAV	CAZ, GEN, SXT	Yes	Septic emboli	D
16	[9]	32/M	IVDU, subcutaneous reservoir	NTV	SXT	No	CHF	D
17	[9]	28/M	IVDU	NAV	CIP, GEN	Yes	Myocardial abscess	C
18	[9]	60/F	Ventriculo-atrial shunt	NTV	TIM, SXT	No	Lung abscess	C
19	[9]	36/M	Dental treatment (3 months)	NAV	TZP, GEN	Yes	CHF	C
20	[9]	69/F	Recent valve replacement (3 months)	PMV, PAV	CAZ, GEN, CIP, SXT	Yes	CHF, persistent bacteremia	D
21	[10]	37/M	Recent mitral valvuloplasty (early)	PMV	CAZ, AMK, CIP then TIM, SXT, COL	Yes	None	C
22	[9]	58/F	Recent valve replacement (3 months)	PMV	SXT	Yes	None	C
23	[9]	62/M	Recent valve replacement (6 months)	PAV	CIP, CHL	No	Aortic dissection	C
24	[9]	40/M	Recent valve replacement (9 months)	PAV	TIM, SXT	No	None	C
25	[9]	44/M	Rheumatic valvular disease	PMV	VAN, GEN	No	Recurrence with septic emboli treated by TMP-SMZ + TOB and surgery	C
26	[11]	44/M	IVDU, HIV, dental treatment, rheumatic aortic and mitral disease	NMV, NAV	LVX, SXT	No	None	C
27	[9]	56/F	CVC	NAV	FEP, CIP, SXT	No	None	C
28	[12]	65/F	NR	PAV	NR	NR	CHF, paravalvular abscess	NA
29	[7]	34/F	Peripheral catheter	PMV	SXT, GEN	No	None	C
30	[13]	38/M	Recent valve replacement (1 year)	PAV	SXT, TIM	Yes	Subannular abscess	C
31	[14]	28/M	Recent valve replacement (3 weeks)	PAV	SXT, CAZ	Yes	None	C
32	[15]	–	Pacemaker	Pacemaker pocket	–	Yes	None	C
33–39	[8]	68–84	Recent valve replacement	PAV	CAZ	Yes	CNS complications in 4	3D
40	[16]	78/F	None	PMV	SXT, CIP, TZP	Yes	Multiple cerebral infarction and paravalvular abscess	D
41	[17]	23/F	Autoimmunity related to SLE					
42	Case 1	35/M	Rheumatic heart disease	Native mitral valve	SXT, LVX	No	None	C
43	Case 2	40/M	Recent valve replacement (4 months)	PMV	VAN, GEN	No	Renal failure	D

Out of 43 cases, 14 patients (33%) died out of various complications.

Abbreviations: AMC, ampicillin; AMK, amikacin; ASD, atrial septal defect; CAR, carbenicillin; CAZ, ceftazidime; CHF, congestive heart failure; CHL, chloramphenicol; CIP, ciprofloxacin; COL, colistin; CVC, central venous catheter; FEP, cefepime; GEN, gentamicin; IVDU, intravenous drug user; KAN, kanamycin; LVX, levofloxacin; MI, myocardial infarction; MOX, moxalactam; NAV, natural aortic valve; NMV, natural mitral valve; NR, not reported; PAV, prosthetic aortic valve; PEN, penicillin; PMV, prosthetic mitral valve; POL, polymyxin; PR, present report; STR, streptomycin; SXT, trimethoprim-sulfamethoxazole; TIC, ticarcillin; TIM, ticarcillin-clavulanic acid; TOB, tobramycin; TZP, piperacillin-tazobactam; VAN, vancomycin; VSD, ventricular septal defect. D, died; C, cure; MM, medical management; SLE, systemic lupus erythematous.

3. Discussion

We share the challenges in managing two cases of *S. maltophilia* endocarditis of different outcomes. One patient was managed successfully with culture-guided early antibiotic therapy, while other case succumbed before the arrival of culture report.

S. maltophilia endocarditis is a rare disease. Only 41 cases have been reported around the world before our observations (Table 1). The clinical features vary from case to case. In-hospital habitat is the reservoir. *S. maltophilia* endocarditis is likely to develop under specific conditions, such as the use of central venous lines, prior cardiac surgery, and intravenous drug abuse.[4–7] Thus, *S. maltophilia* from contaminated medical equipment in hospitals may cause endocarditis when the skin barrier is broken.[2,6] In particular, prior valve replacement is one of the predisposing factors that accounts for approximately 40–60% of the endocarditis cases.[5–7]

Treatment of endocarditis caused by *S. maltophilia* comprises appropriate antibiotic therapy and removal of indwelling infected foreign material in the body. Because of limited experience and resistance to multiple antibiotics, the treatment is purely based on consensus and regional culture sensitivity pattern. *S. maltophilia* is resistant to penicillin, cephalosporin, and Carbapenems. Sulfamethoxazole-trimethoprim is selected as the first-line antibiotic, supported by in vitro susceptibility.[2,3] Since sulfamethoxazole-trimethoprim is bacteriostatic against the most isolates, it is used in combination with other antibiotics for synergistic effect. The difficulty in treating *S. maltophilia* endocarditis with antibiotic therapy arises due to sulfamethoxazole-trimethoprim intolerance. Several reports have stated that combination therapy with fluoroquinolones, aminoglycosides, and 3rd or 4th generation cephalosporin is effective.[6,8] Discrepancies between the in vitro susceptibility data and clinical outcome have been noted in the case of *S. maltophilia* infections.[1,3] Both morbidity and mortality rates are high in cases of endocarditis caused by *S. maltophilia*. The overall incidence of mortality is approximately 34.8% (15/43), as has been summarized from case reports around the world (Table 1). Complications such as cerebral vascular disease, congestive heart failure, and organic abscess are seen in 70–80% of patients[5–7] because of antibiotic resistance. Autoimmunity could be included as a novel predisposing factor for *S. maltophilia* endocarditis, as reported in case report by Carrillo-Córdova et al.[17] Until now, to the best of our knowledge, there is no published report of infective endocarditis caused by *S. maltophilia* from Indian subcontinent.

4. Conclusion

The true epidemiological profile of infective endocarditis due to *S. maltophilia* is emerging. The treatment for this organism has not been addressed in most recently updated infective endocarditis guidelines. The very reason may be its rare occurrence and paucity of experience. It is important to identify this microorganism as quickly as possible, since *S. maltophilia* is resistant to first line antibiotic therapy generally used in case of nosocomial infections. This case report reemphasizes the meticulous steps in the prevention of device-related infections in operation theaters and cardiac catheterization laboratories. These are the first two case reports of infective endocarditis caused by *S. maltophilia* from India.

Conflicts of interest

The authors have none to declare.

REFERENCES

1. Denton M, Kerr KG. Microbiological and clinical aspects of infection associated with *Stenotrophomonas maltophilia*. *Clin Microbiol Rev*. 1998;11:57–80.
2. Dignani MC, Grazziutti M, Anaissie E. *Stenotrophomonas maltophilia* infections. *Semin Respir Crit Care Med*. 2003;24:89–98.
3. Looney WJ, Narita M, Muhlemann K. *Stenotrophomonas maltophilia*: an emerging opportunist human pathogen. *Lancet Infect Dis*. 2009;9:312–323.
4. Munter RG, Yinnon AM, Schlesinger Y, Hershko C. Infective endocarditis due to *Stenotrophomonas (Xanthomonas) maltophilia*. *Eur J Clin Microbiol Infect Dis*. 1998;17:353–356.
5. Khan IA, Mehta NJ. *Stenotrophomonas maltophilia* endocarditis: a systematic review. *Angiology*. 2002;53:49–55.
6. Crum NF, Utz GC, Wallace MR. *Stenotrophomonas maltophilia* endocarditis. *Scand J Infect Dis*. 2002;34:925–927.
7. Bayle S, Rovery C, Sbragia D, Brouqui P. *Stenotrophomonas maltophilia* prosthetic valve endocarditis: a case report. *J Med Case Reports*. 2008;2:174.
8. Muller-Premru M, Gabrijelcic T, Gersak B, et al. Cluster of *Stenotrophomonas maltophilia* endocarditis after prosthetic valve replacement. *Wein Klin Wochenschr*. 2008;120:566–570.
9. Lopez RR, Lado Lado FL, Sanchez A, et al. Endocarditis caused by *Stenotrophomonas maltophilia*: report of a case and review of literature. *Ann Med Interna*. 2003;20:312–316.
10. Wu PS, Lu CY, Chang LY, et al. *Stenotrophomonas maltophilia* bacteremia in pediatric patients—a 10-year analysis. *J Microbiol Immunol Infect*. 2006;39:144–149.
11. Senol E. *Stenotrophomonas maltophilia*: the significance and role as a nosocomial pathogen. *J Hosp Infect*. 2004;57:1–7.
12. Mermel LA, Farr BM, Sherertz RJ, et al. Guidelines for the management of intravascular catheter-related infections. *Clin Infect Dis*. 2001;32:1249–1272.
13. Ucak A, Goksel OS, Inan K, et al. Prosthetic aortic valve endocarditis due to *Stenotrophomonas maltophilia* complicated by sub-annular abscess. *Acta Chir Belg*. 2008;108:258–260.
14. Sanioğlu S, Sokullu O, Yavuz SS, Kut MS, Palaz FK, Bilgen FS. *Stenotrophomonas maltophilia* endocarditis treated with moxifloxacin-ceftazidime combination and annular wrapping technique. *Anadolu Kardiyol Derg*. 2008;8:70–80.
15. Takigawa M, Noda T, Kurita T, et al. Extremely late pacemaker-infective endocarditis due to *Stenotrophomonas maltophilia*. *Cardiology*. 2008;110:226–229.
16. Katayama T, Tsuruya Y, Ishikawa S. *Stenotrophomonas maltophilia* endocarditis of prosthetic mitral valve. *Intern Med*. 2010;49:1775–1777.
17. Carrillo-Córdova JR, Amezcua-Guerra LM. Autoimmunity as a possible predisposing factor for *Stenotrophomonas maltophilia* endocarditis. *Arch Cardiol Mex*. 2012;82:204–207.