



Case report

Endocarditis caused by *Stenotrophomonas maltophilia*—A rare presentation of an emerging opportunistic pathogen



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ARTICLE INFO

Article history:

Received 18 April 2019

Received in revised form 7 May 2019

Accepted 7 May 2019

Keywords:

S. Maltophilia

Endocarditis

ABSTRACT

First isolated in 1943, *Stenotrophomonas maltophilia* (*S. maltophilia*) has historically been of little significance as it was considered a pathogen of low virulence noted to rarely infect immunocompromised hosts. However, over the last 30 years the prevalence of infection caused by the organism has increased significantly. Bacterial endocarditis from *S. maltophilia* remains exceedingly rare with only a small number of reported cases in the literature. This case involves a 27 year old male with sickle cell anemia with an indwelling right subclavian port who presented to the emergency department with myalgias, fever, and chills. His initial blood cultures grew Gram negative rods later identified as *S. maltophilia*. Transthoracic echocardiogram showed a mass in the right atrium. Transesophageal echocardiogram revealed a large C-shaped mass with attachment to the tricuspid annulus, mitral valve wall, and port tip in right atrium. The patient underwent sternotomy with removal of the vegetation to prevent embolization. He was treated with intravenous ciprofloxacin and oral trimethoprim/sulfamethoxazole to complete a full 6 weeks of therapy, making a full recovery. This report will further explore the unique presentation of this pathogen along with its epidemiology, resistance mechanisms, risk factors for infection, diagnosis, and appropriate antimicrobial treatment.

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Introduction

First isolated in 1943, *Stenotrophomonas maltophilia* (*S. maltophilia*) has historically been of little significance as it was considered a pathogen of low virulence noted to rarely infect immunocompromised hosts [1]. However, over the last 30 years the prevalence of infection caused by the organism has increased significantly [2]. It has simultaneously demonstrated resistance to many common antimicrobials via multiple mechanisms, making early identification of this lethal pathogen extremely important. Bacterial endocarditis from *S. maltophilia* remains exceedingly rare with only a small number of reported cases in the literature.

We present here a very rare case of *S. maltophilia* endocarditis. This case highlights an opportunistic pathogen that can present in a number of different ways. Through a comprehensive literature review, this organism was isolated as the cause of endocarditis in only 43 cases so far.

Case report

A 27 year old male with a past medical history of sickle cell anemia requiring frequent admissions for sickle cell crisis, with a right subclavian port placed two years prior for frequent blood transfusions, presented to the emergency department with generalized myalgias, weakness, fever and chills for two days duration. The patient denied any chest pain, cough, vomiting, or diarrhea. Vitals upon presentation were significant for a fever of 100.8° Fahrenheit, heart rate of 123 beats per minute, blood pressure of 146/81 mmHg, and an oxygen saturation of 99% on room air. His lungs were clear to auscultation, cardiovascular exam was unremarkable, and the port site was non-erythematous and non-tender. Initial labs were significant for anemia with a hgb of 6.9, an elevated reticulocyte count of 17.6, and a leukocytosis of 14,000. Chest X-ray was grossly unremarkable.

The patient was initially treated for sickle cell crisis with intravenous fluids, pain control and hydroxyurea. Two sets of blood cultures were also collected. On the second day of hospitalization both sets of blood cultures were growing gram negative bacilli. Patient was started on cefepime while the organism was speciated and Infectious Disease service was consulted. Two additional sets of blood cultures were drawn (one peripherally and one from the

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port-a-cath). These also grew Gram negative bacilli. The bacteria was identified as *S. maltophilia*, susceptible to trimethoprim/sulfamethoxazole (TMP/SMX) and levofloxacin. Antimicrobial therapy was promptly changed to IV TMP/SMX but was later changed to levofloxacin 1 g daily (higher dose based on the minimum inhibitory concentration) due to hyperkalemia.

Transthoracic echo was preformed and revealed an echogenic density which was suspicious for a thrombus or mass. Transesophageal echo then confirmed the presence of a large (2 cm × 3.9 cm) C-shaped independently mobile mass, attached to the right atrial wall with attachment to the tricuspid annulus and possibly the port tip identified within the atrium. The port was removed successfully and sent for culture which also grew *S. maltophilia*. Cardiothoracic surgery was consulted for surgical intervention. At the time of surgical consultation, the patient had cleared blood cultures, but given the size of the mass, its mobility, and risk of embolization the decision was made for surgical intervention. The patient underwent sternotomy with excision and culture of the mass without complication. Tissue culture was negative for growth in setting of appropriate antimicrobial therapy for 14 days leading up to surgery. Histopathology of the mass revealed many neutrophils without epithelial cells or organisms. Follow-up transthoracic echo showed only mild tricuspid regurgitation (Figs. 1–4).

A peripherally inserted central catheter was placed prior to discharge and the patient was discharged with a 6 week course of intravenous ciprofloxacin plus oral TMP/SMX. The patient was seen and evaluated in the clinic 2 weeks later and was on his way to a full recovery.

Discussion

Over the last 30 years, *S. maltophilia* has emerged as an important cause of nosocomial infections. ¹ *S. maltophilia* is a gram negative, aerobic, non-glucose fermenting bacillus [1]. It is generally smaller than the other species within the same genus and it is mobile due to a polar flagellum [2]. It is quite ubiquitous in nature, thriving in humid and aquatic areas being found most commonly in rivers, lakes, soil, plants, shower heads, and faucets [6]. In the hospital setting it can survive on any humid surface such as tubing, catheters, hemodialysis equipment, ventilators, and endoscopes [1]. Although generally thought of as a low virulence organism it can cause serious problems for immunocompromised patients due to multiple virulence factors and extensive resistance to antimicrobials.

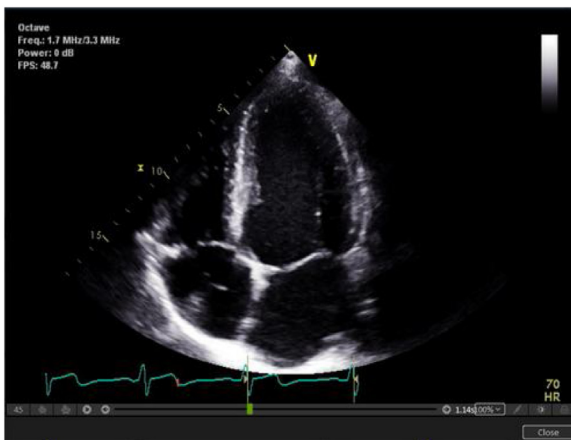


Fig. 1. Transthoracic echocardiogram Apical four chamber view demonstrating thrombus attached to superior right atrial wall and revealing port tip present in atrial chamber.

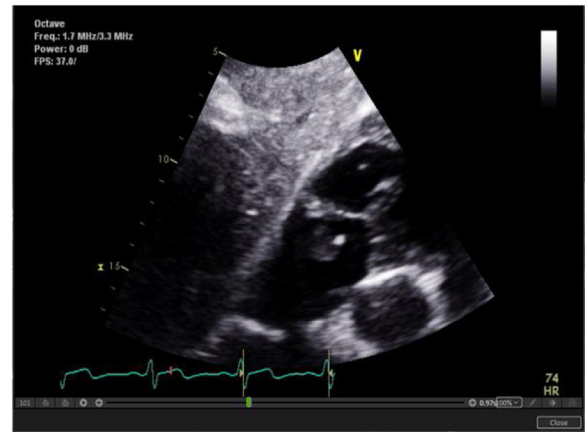


Fig. 2. Transthoracic echocardiogram: parasternal short axis view showing thrombus attached to tricuspid valve.

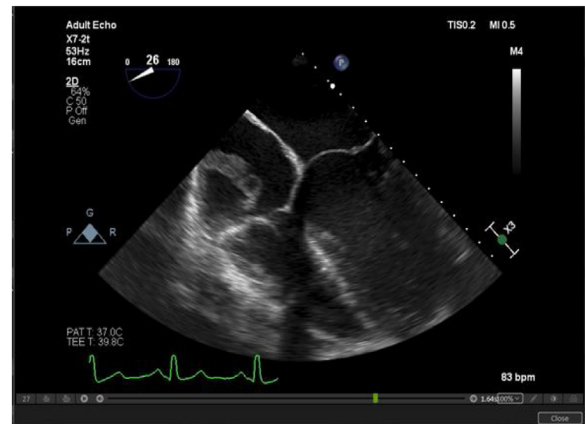


Fig. 3. Transesophageal echocardiogram: Four chamber view, reveals attachment of thrombus to atrial wall.



Fig. 4. Transesophageal echocardiogram: Bicaval view with measurement of thrombus 3.9 × 2.0 cm in size.

Although generally rare, the prevalence of *S. maltophilia* infections has gradually increased, with studies showing a prevalence of 0.8%–1.4% of documented infections from 1997 to 2003 compared to a prevalence of 1.3%–1.68% infections from 2007 to 2012 [1]. This increase in prevalence is likely due to multiple factors such as an aging population, increase in prevalence of

immunocompromised hosts, development of multiple resistance factors to antimicrobials, and the use of invasive interventions in health care. However, even though the overall rate of infection by this pathogen has increased, *S. maltophilia* endocarditis still remains an exceedingly rare diagnosis. As of 2016, there have only been 43 confirmed cases of *S. maltophilia* endocarditis [3]. *S. maltophilia* infection generally presents as bacteremia or an upper respiratory infection [4]. As it pertains to endocarditis, the most significant risk factors included use of a central venous catheter, prior cardiac surgery, intravenous drug use, and prior valve replacement (accounting for 40–60% of reported cases) [3]. Early diagnosis is key in the successful treatment of this infection as mortality in *S. maltophilia* endocarditis is high (34.8%) [3]. The diagnosis should be made by early blood cultures. Once *S. maltophilia* bacteremia has been established, imaging via trans-thoracic and/or transesophageal echocardiogram should be performed in for further evaluation for endocarditis.

S. maltophilia endocarditis represents a clinical challenge when it comes to antimicrobial choice due to the rarity of the disease, extensive antimicrobial resistance, and the lack of uniform guidelines regarding treatment [3]. *S. maltophilia* exhibits high antimicrobial resistance due to both inherent and acquired genes, which is particularly dangerous as it means resistance to individual antimicrobials could broaden [2]. *S. maltophilia* has been found to possess B-lactamase activity via L1-L2 proteins, efflux pump genes such as SmeABC, SmeDEF, and aminoglycoside acetylsynthase, among others [1,2]. These mechanisms have made most strains of the disease resistance to most penicillins, cephalosporins, aminoglycosides, and carbapenems. Although there is no consensus, the preferred option in treatment seems to be TMP/SMX [1–8]. Most studies have shown a resistance rate of around 10% or less [1] although others have observed a rate closer to 29% leading to recommendations and studies involving TMP/SMX dual therapy with fluoroquinolones or tetracyclines [6]. If TMP/SMX is not tolerated, levofloxacin is commonly thought of as an appropriate alternative. However recent studies have shown a decrease in sensitivity from 83 percent to 77 percent [1]. Other antimicrobials that have been used in case reports include ceftazidime, minocycline, doxycycline, polymyxin, and fosfomycin [1–8].

Although these antimicrobials have been studied with some success, the most important step in treatment is early obtainment of sensitivities in order to appropriately focus treatment. Misdiagnosis or late diagnosis of *S. maltophilia* infection can lead to development of fatal complications. In a number of cases, the use of prolonged broad-spectrum antimicrobial therapy (such as carbapenems to which *S. maltophilia* is naturally resistant) resulted in a rapid colonization [2]. In addition to antimicrobial therapy, in cases of endocarditis it is imperative that any indwelling catheter

be removed. Surgical consultation must be obtained for evaluation if vegetations are noted on echocardiography. Failure to seek appropriate surgical treatment can result in persistent bacteremia, risk of embolization, and heart failure with valvular involvement.

Conclusion

This case highlights an emerging nosocomial pathogen that presented in a rare way. It demonstrates the importance of early culture and sensitivity given the extensive resistance pattern *S. maltophilia* exhibits. Given the high mortality and morbidity of *S. maltophilia* endocarditis, more research is needed in order to establish effective guidelines for the treatment of this potentially devastating disease.

Disclosure

The Authors state that the views expressed in the submitted article are their own and not an official position of the institution or funder.

Conflict of interest

No relevant conflict of interests exist.

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