Early diagnosis and prompt management holds the key for successful outcome. All efforts should be made to correct the underlying and predisposing factor. Surgical debridement should be done at the earliest to remove maximum amount of devitalised tissue that is possible. Intravenous amphotericin B (liposomal) is the main stay of therapy. It is given at a dose of 5-10 mg/kg/day and the duration depends on the clinical response. A dose of at least 2 g is necessary in most cases.[6] Posconazole[12] has emerged as an alternative salvage therapy with good clinical response. Diagnosis at the early stage of the disease is pivotal as mortality rate of 94% is seen among patients who develop disseminated disease as a consequence to primary cutaneous disease.[1]

Conclusion

Our case further strengthens and establishes Syncephalastrum as an aetiologic agent for zygomycosis. Mucuromycosis represent an uncommon but life-threatening infection in immuno-compromised, especially diabetics. Prompt treatment that includes correction of underlying condition, surgical debridement and intensive antifungal therapy form the cornerstone of successful management. Untreated, these agents can cause highly invasive and fatal disease. Patients having diabetes, organ transplants, haematopoetic stem cell transplant and AIDS are especially prone for contracting these diseases. The possibility of these atypical infections should always be borne in mind while dealing with the above-said patient groups.

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Sphingomonas paucimobilis bacteraemia and shock in a patient with rheumatic carditis

***Y Yozgat, A Kilic, C Karadeniz, R Ozdemir, O Doksoz, G Gulfidan, T Mese**

Abstract

Acute rheumatic fever (ARF) carditis is treated with steroids, which can cause changes in the cellular immune response, especially decreased CD3 (+) T cells. Nosocomial infections due to steroid use for treatment of ARF carditis or secondary to the changes in the cellular immune response have not been reported in the literature. *Sphingomonas paucimobilis* is a Gram-negative bacillus causing community- and hospital-acquired infections. It has been reported as causing bacteraemia/sepsis, pneumonia or peritonitis in patients with malignancies, immunosuppression or diabetes. We

present a case with *S. paucimobilis* bacteraemia/sepsis and shock after administration of steroids for treatment of ARF carditis. We suggest early identification of the causative agent and appropriate adjustments of the treatment plan to avoid shock and possible mortality. This is the first reported case of *S. paucimobilis* bacteraemia/sepsis in the setting of steroid use for ARF carditis.

Key words: *Acute rheumatic fever, bacteraemia, shock, Sphingomonas paucimobilis*

Introduction

Acute rheumatic fever (ARF) is a multisystemic disorder seen after group A streptococcal pharyngitis, which is frequently encountered in children and adolescents between the ages of 5 and 15. ARF carditis has been reported in 35-50% of patients with ARF.[1] In cases with ARF, changes in cellular immune response, especially decreased quantities of CD3 (+) T cells, have been reported. Nosocomial infections secondary to steroid use during treatment of ARF carditis or changes in the cellular immune response have not yet been reported.

Sphingomonas paucimobilis (formerly referred to as *Pseudomonas paucimobilis*) is a yellow pigment-forming, aerobic, non-fermentative, non-spore-forming, oxidase and catalase-positive Gram-negative bacillus.[2] The bacterium is commonly found in nature, particularly in soil and drinking water and on the surface of plants; in the hospitals it has been isolated in haemodialysis instruments, humidifiers, water containers, distilled water tanks, basins and thermometer probes.[3]

S. paucimobilis has recently gained importance in terms of hospital-acquired infections, being associated with bacteraemia/sepsis, pneumonia or peritonitis in patients with malignancies, immunosuppression or diabetes.[4] However, *S. paucimobilis* bacteraemia/sepsis in patients with ARF have not yet been reported.

We present a case diagnosed with bacteraemia of *S. paucimobilis* and shock developing in the presence of ARF carditis with CD3 (+) T-cell reduction. To the best of our knowledge, this is the first such report in the literature.

Case Report

A 13-year-old female patient was admitted with complaints of sore throat, mild fever, malaise and fatigue. She was in good general condition, conscious, cooperative and mildly dehydrated. Body temperature was 37.7°C, pulse rate 124 beats per minute, blood pressure 105/60 mmHg and respiratory rate 18 per minute. History revealed a severe episode of pneumonia and pleurisy requiring chest tube insertion, prolonged hospitalisation and intravenous antibiotics a year ago. The causative agent had not been identified and treated empirically with vancomycin and ceftriaxone. Electrocardiography was normal. Mild mediastinal expansion and cardiomegaly was observed on chest X-ray [Figure 1]. Echocardiography showed an ejection fraction of 68%, fractional shortening 34%, second-degree mitral and tricuspid regurgitation, mild aortic and pulmonic regurgitation. Pericardial echogenicity was increased with moderate fluid accumulation [Figure 2]. Biochemical analyses were as follows: White blood cell: 10700/mm^3, haemoglobin: 9.7 g/dl, MCV: 80 fl, platelet: 491.000/mm^3, ESR: 117 mm/h, CRP: 11.8 mg/dl and ASO: 769.00 IU/ml. Differential count showed 88% polymorphonuclear leukocytes, 10% lymphocytes and 2% monocytes; there were no atypical cells. Blood glucose, liver and kidney function tests, electrolytes, uric acid, LDH and lipid levels were normal, as were blood gas analysis, cardiac panel, ANA, anti-dsDNA, salmonella and Brucella group agglutination tests and ferritin levels. Urinalysis was unremarkable. Three consecutive blood samples were obtained hourly for culture.

As the patient had one major (carditis) and two minor criteria (elevated ESR, CRP and temperature) along with ASO elevation, a diagnosis of ARF carditis and pericardial effusion was made. For primary eradication of GAS, 1.200.000 units of benzathine penicillin were administered intramuscularly. As she was hypotensive, dopamine (5 mcg/kg/min), spironolactone (2 mg/kg/day), prednisolone (2 mg/kg/day) and ceftriaxone (75 mg/kg/day) were added.

On the third day of hospitalisation, her general condition suddenly deteriorated into a clinical picture of shock. The treatment plan was modified to include dopamine, dobutamine, norepinephrine and fluids.

In the fourth day, the general condition of the patient was slightly better. Gram-negative bacilli were isolated from all of the blood cultures taken at the time of admission.

S. paucimobilis was detected with standard aerobic and anaerobic blood culture media in automated blood culture systems (BacT/ALERT PF Plus, bioMérieux, France). Identification and antibiotic susceptibility testing of the isolate were done with the VITEK 2 Compact system (bioMérieux, France). The results were interpreted using Clinical and Laboratory Standards Institute (CLSI) standards. Sensitivity results were reported as sensitive or resistant based on CLSI

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criteria. The strain was found to be resistant to gentamicin, amikacin, sulbactam + ampicillin and sensitive to meropenem, ceftazidime and piperacillin. Accordingly, ceftriaxone was withdrawn and meropenem 60 mg/day was started. Immunoglobulins and lymphocyte panel were studied. As the percentage of CD3 (+) T cells were lower [Table 1], CD3 TRC complex chain was considered to be defective and 0.4 g/kg intravenous immunoglobulin was given.

The clinical status and biochemical analyses gradually improved after the fifth day; ESR dropped to 60 mm/h and CRP to 1.63 mg/dl. Control blood cultures were reported negative. She was discharged after 10 days of hospital stay with usual ARF outpatient treatment plan with oral prednisolone and subsequent acetyl salicylic acid. Three months after the discharge, her general condition was good, percentage of CD3 (+) T cells were normal, minimal aortic and mitral regurgitation was detected with echocardiography.

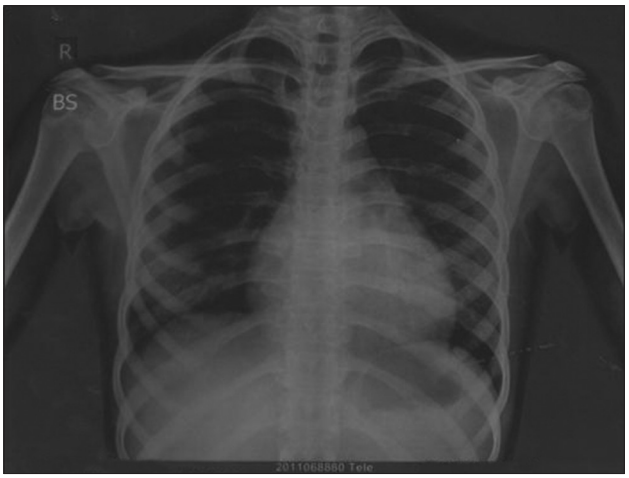

Figure 1: Posteroanterior chest X-ray showing mild cardiomegaly and mediastinal expansion

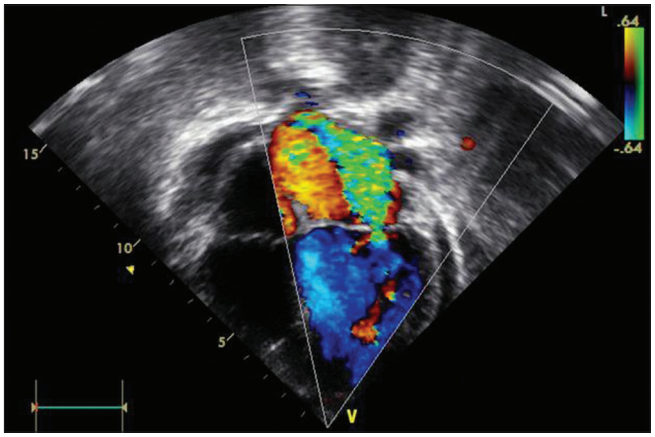

Figure 2: Echocardiography showing second-degree mitral regurgitation and pericardial effusion of 10 mm around left ventricle

Table 1: The percentages of CD3, CD4, CD8 T cells, CD4/CD8 T cell ratios and CD19 B cells in the patient

Lymphocyte subset panel	CD 3	CD 4	CD 8	CD4/ CD8	CD 16-56	CD 19
Patient, %	37.7	29.9	20.7	1.2	34.5	30.7
(normal range, %)	(60-85)	(22-59)	(14-48)	(0.9-1.4)	(6-37)	(7-28)

Discussion

Patients with ARF carditis are treated with steroids and subsequent acetyl salicylic acid. Changes in the cellular immune response, especially CD3 (+) T-cell reduction, have been reported in patients with ARF.[5] Our case was given steroids for severe ARF carditis and reduced numbers of CD3 (+) T-cell reduction was identified.

S. paucimobilis is an aerobic, non-fermentative Gram-negative bacillus with slow motility found commonly in nature, particularly in soil and drinking water. In the hospitals, it has been isolated in haemodialysis instruments, humidifiers, basins and thermometer probes.[2,3] In a meta-analysis published by Ryan and Adley, bacteraemia/sepsis and peritonitis were reported as the most common clinical forms.[6]

The origin of nosocomial *S. paucimobilis* infection may be endogenous (colonisation secondary to the previous infection) or environmental (catheters, infected distilled water, haemodialysis fluids).[7] The fact that all of the blood samples obtained at admission was positive for *S. paucimobilis* in our case makes us consider that the organism had been endogenous, possibly secondary to pleurisy and chest tube insertion a year ago.

In another review of 16 cases of *S. paucimobilis* bacteraemia, the underlying diseases were reported as diabetes in 11.9%, immunosuppressive therapy in 40.5% and Malignancy in 57.1% of cases.[4] In our case, we consider that *S. paucimobilis* was present due to the previous pleural infection, and the suppression was removed with the steroid therapy causing CD3 (+) T-cell reduction leading to bacteraemia and shock.

S. paucimobilis is a bacterium with low virulence and a rare cause for life-threatening infections, possibly due to the lack of lipopolysaccharides in the cell wall, rendering it resistant to the effects of endotoxins.[8] *S. paucimobilis* is generally sensitive to imipenem, tetracycline, chloramphenicol, cotrimoxazole and carbapenem and aminoglycosides. Its sensitivity to third-generation cephalosporins and quinolones is variable.[9] Our case initially developed shock, but rapidly improved after the administration of meropenem.

To the best of our knowledge, this is the first case of coexisting ARF carditis, *S. paucimobilis* bacteraemia/sepsis and shock reported in the literature. In patients with ARF carditis,

S. paucimobilis bacteraemia should be considered when sepsis and shock develop during corticosteroid therapy and antibiotic sensitivity should be assessed for appropriate treatment.

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