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Case Report

Stenotrophomonas Maltophilia/S. Maltophilia Sepsis Presenting with Perianal Cellulitis and Pneumonia in a Leukemic Child

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ABSTRACT

Infections caused by *Stenotrophomonas maltophilia* may be fatal in diseases such as leukaemia which causes immunodeficiency. Herein, we report a rare case of 8-year-old girl with acute lymphoblastic leukaemia, who developed perianal cellulitis and pneumonia secondary to *S. maltophilia* bacteraemia while undergoing broad spectrum antimicrobial agents for neutropenic fever.
Keywords: *S. maltophilia*, cellulitis, pneumonia, leukaemia

Introduction

Stenotrophomonas maltophilia infections are increasingly seen in immunocompromised patients, as it is an opportunistic pathogen with low virulence (1). Risk factors for *S. maltophilia* infection include prolonged hospitalization in intensive care units, medical devices, use of broad-spectrum antibiotics and malignancy (1,2). Infections are more prevalent during severe neutropenic periods, and in cases in which immunosuppression develops; however, its mode of transmission is not clear. It is thought to invade from damaged mucous membranes and central venous catheters through colonization (2). This case emphasizes how difficult it is to distinguish between invasive bacterial infections and fungal infections in patients with severe neutropenia, as well as that skin infections can be associated with *S. maltophilia*.

Case Report

An eight-year-old girl was admitted to the hospital because of fever, bone and joint pain for 3 months. Physical examination revealed paleness, painful movement in the right hip joint and lack of organomegaly. Laboratory studies showed leukocytosis of 93.000/mm^3, hemoglobin of 9.9 g/dL, and a platelet count of 117.000/mm^3. A bone marrow aspiration established a diagnosis of acute lymphoblastic leukaemia (ALL), and chemotherapy was started (ALL IC-BFM 2009). On the 15th day of induction therapy, a Hickman catheter was inserted to act as a venous catheter. On the 55th day of induction therapy, she developed severe neutropenia. On the 60th day of induction chemotherapy, her body temperature increased to 39 °C. A complete blood count showed a white blood cell count of 300/mm^3, an absolute neutrophil count of 0, and C-reactive protein

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(CRP) 0.31 mg/dL (normal range 0-0.8). Therefore, she was considered to have febrile neutropenia at that time. In accordance with the clinical practice guideline for the use of antimicrobial agents in neutropenic patients with cancer: 2010 update by the Infectious Diseases Society of America guidelines, antibiotic therapy was initiated with carbapenem, aminoglycoside, and vancomycin. Her fever continued for 5 days CRP: (15.5 mg/dL), and so Liposomal Amphotericin B was added to the therapy. On day 7 of febrile neutropenia, physical examination revealed a hyperaemic, tender, irritable and non-fluctuating lesion in the perianal region, which was seen as cellulitis. Its size increased to 5x6 cm. The pelvic magnetic resonance imaging revealed a suprasphincteric and superficial perianal fistula in the right perianal region (Figure 1). The biopsy from her lesion couldn't be done because of bacteremia risk from the infected area due to severe neutropenia. Due to persistent fever, carbapenem was stopped and colistin initiated. Galactomannan antigen test result was positive with a level of 2.3 on the 9th day of febrile neutropenia. Since invasive aspergillosis could not be ruled out, voriconazole was added. Four days after the development of the lesion in the perianal region, pain started

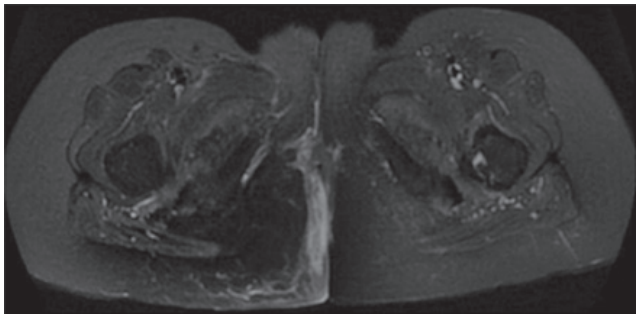

Figure 1. Pelvic magnetic resonance imaging showed a cellulitis in the right perianal region

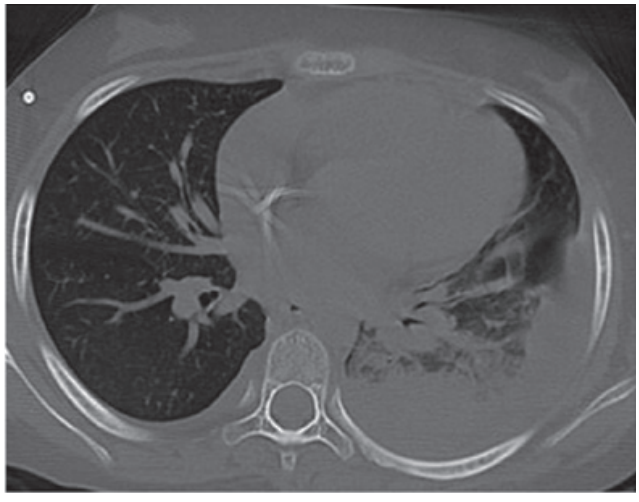

Figure 2. Thorax computed tomography showed a pneumonic consolidation and pleural effusion in the left lower pulmonary lobe

in the left side of the chest. Thorax computed tomography revealed pneumonic consolidation and pleural effusion in the lower left lobe (Figure 2). Her galactomannan antigen titre began to decrease 4 days later (0.9). Catheter cultures were negative. *S. maltophilia* was isolated in two blood cultures. Trimethoprim-sulfamethoxazole (15 mg/kg/day, parenterally) was initiated. According to culture antibiogram results, ciprofloxacin was also added. Consolidation developed in the upper right lobe of the lung, subsequently. Within a few days, the patient developed respiratory failure. Despite aggressive supportive therapy, the patient's condition deteriorated and she died 7 days later.

After receiving informed consent, a lung necropsy was performed, two from the left lung and one from the right lung. Pathological investigation revealed no fungal spores or hypha; massive intra-alveolar haemorrhage was also detected. Post-mortem catheter and blood cultures were negative. Written informed consent was obtained from the patient's parents.

Discussion

Patients with haematological disorders, particularly leukemic patients, are in a high-risk group for *S. maltophilia* infections, due to chemotherapy-associated neutropenia and immunodeficiency (2). Use of wide-spectrum antibiotics and long-term catheterization increases this risk (3). Our patient had an underlying disease of ALL as a haematological malignancy, and was on immunosuppressive treatment. A history of hospitalization for 60 days, with wide-spectrum antibiotics for 10 days for neutropenic fever, must be noted. In addition, the patient had a central venous catheter and multiple risk factors for *S. maltophilia*.

S. maltophilia is associated with an increasing number of infections at various sites, particularly pneumonias, bacteremias and cellulitis (4) In patients with severe immunodeficiency, it is described as a haemorrhagic pneumonia pathogen, and does not have characteristic radiographic features that can be differentiated from other bacterial pneumonia. Its radiologic appearance may exhibit a unilateral or bilateral pattern, but it is rarely accompanied by pleural effusion (4). The clinical findings resemble those of pulmonary aspergillosis (5). Generally, cough and dyspnea frequently accompany it, which may manifest with pleuritic chest pain and haemoptysis (6). Our patient had chest pain symptoms, although radiological investigation revealed no specific finding as the causative agent.

Galactomannan is the polysaccharide, which is the building block of the cell wall of aspergillosis and other types of fungus. In some patients, clinical signs and symptoms of invasive aspergillosis can be demonstrated before appearing. False positive results can also occur due to the colonization of the airways by the aspergillosis species (7). In our patient,

we did not see a test result to support fungal infection, other than galactomannan positivity. A low galactomannan titre reduced the possibility of a fungal infection diagnosis, while the positive galactomannan antigen test was thought to be aspergillosis colonization of the respiratory tract.

The most remarkable type of soft tissue infection due to *S. maltophilia* is the metastatic cellulitis, primary cellulitis, infected mucocutaneous ulcers and ecthyma gangrenosum (1,8-10). A study of 114 patients with malignancies and *S. maltophilia* infection reported metastatic cellulitis in 6 patients, primary cellulitis in 5 patients and infected mucocutaneous ulcers in six patients (1). Moser et al. (8) reported skin infections in three of 13 neutropenic patients with *S. maltophilia* bacteremia. They reported tender, erythematous, warm subcutaneous nodules, suggesting cellulitis in one patient, tender, red, warm, poorly-demarcated subcutaneous lesions resembling cellulitis in another, and well-demarcated skin lesions resembling cellulitis in a third patient. Mucocutaneous infection by *S. maltophilia* consisted of infected ulcers of the gingiva, lips and buccal mucosa. So far, 10 cases were reported, including 4 cases which were associated with metastatic lesions (1,8,10).

In conclusion, *S. maltophilia* is an opportunistic pathogen with a high mortality rate in immunocompromised patients. Rarely, it may cause skin infections, such as subcutaneous nodules and metastatic cellulitis. In our patient, colonization of bacteria through the fistula area led to a breach of the mucosal barrier in the anal region, and may therefore have caused bacteremia and pneumonia. A long-term history of hospitalization, persistent fever (despite broad-spectrum antibiotics), pneumonia and skin infections associated with haematological malignancy in patients with *S. maltophilia* infection must be considered.

Ethics

Informed Consent: Consent form was obtained from the patient's parents.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Concept: Z.C.Ö., A.B.T., Design: Z.C.Ö., Ö.B., Data Collection or Processing: Z.C.Ö., Analysis or Interpretation: Z.C.Ö., Ö.B., A.B.T., Literature Search: Z.C.Ö., A.B.T., Writing: Z.C.Ö.

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