CASE REPORT

CUTANEOUS MELIOIDOSIS AND NECROTIZING FASCIITIS WITH PULMONARY INVOLVEMENT IN A CHICKEN SELLER

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Abstract. Melioidosis is endemic in Malaysia. Cutaneous melioidosis is one manifestation and it may progress to necrotizing fasciitis. The case highlights a 46-year old male, a chicken-seller who presented with scalp cellulitis which later progressed to necrotizing fasciitis and pneumonia are presented here. It illustrates several key features of the presentation, prompt laboratory diagnosis and early treatment of melioidosis which saved the patient's life.

INTRODUCTION

Melioidosis is endemic in Southeast Asia, and Malaysia is not an exception. A study in Malaysia revealed pneumonia as the most common presenting feature and diabetes mellitus as the most common underlying risk factor with mortality rates as high as 65% (Puthucheary *et al*, 1992). Cutaneous melioidosis with necrotizing fasciitis however, is rare. A case of cutaneous melioidosis with pulmonary infection and necrotizing fasciitis in a man who encountered this disease through his occupation as a chicken-seller is presented.

CASE REPORT

A 46-year old chicken seller was admitted with fever and cough for 10 days. He gave a history of a localized skin lesion over the occipital region which had been present for 2 weeks. He was a diabetic but was non-compliant with his medications. Clinical examination revealed a fever of 40°C, pulse rate 110/ min, and blood pressure 150/90/mmHg.

Tel: +603 55442850; Fax: + 603 55442831 E-mail: arizaadnan@hotmail.com Local examination of the occipital region revealed the presence of bullae over a reddened area of 10 x 10 cm. Crepitations was audible over the right lung. Chest radiograph revealed multiple opacities with cavitations in both lung fields. Based on these findings, penicillin and cloxacillin were commenced. Laboratory investigations revealed a blood sugar of 27.1 mmol/l and a total white count of 18.9 x 10⁹/l.

Over the next 48 hours, he continued to have spiking fever and appeared toxic. At this point, the microbiology laboratory alerted the doctor that the blood and sputum cultures which were taken on the day of admission were suspicious for Burkholderia psuedomallei. A diagnosis of meliodosis was then considered. He was then started on ceftazidime. His general condition however, remained poor despite the antibiotics. He was breathless, hypoxemic, was obtunded and needed oxygen support via ventimask. Doxycycline and trimethoprimsulphamethoxazole were added when the culture and sensitivity results confirmed the diagnosis of melioidosis. The scalp infection had then spread to the neck. Immediate surgical intervention was undertaken. He underwent incision and drainage, followed by debridement of the lesion which comprised a large area of skin necrosis with bullae formation with

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serous and pus. The result of the pus culture yielded *Burkholderia pseudomallei*.

One week following the onset of treatment, the fever began to improve. Regular debridement and wound care were carried out on the scalp infection. He subsequently underwent a skin graft for the scalp lesion. He received a 17-day course of parenteral ceftazidime and was discharged with oral trimethoprimsulphamethoxazole and doxycyline as maintenance therapy.

DISCUSSION

In this case, the patient, had defined risk factors. He was a diabetic and his job involved handling and slaughtering chickens. This predisposed him to contaminated soil (found on the surface of the chickens) which harbored the infective agent. One cannot exclude the scalp as the primary site of infection. Reports of the mode of acquisition suggested the causative organism, Burkholderia pseudomallei, exists as a free-living saprophyte and has been cultivated from stagnant water and soil. Patients are infected by direct penetration of the organism through minor skin abrasions or by inhalation of dust. Inoculation is the most likely mode of acquisition for this patient through minor skin abrasions during his contact with chickens. Minor wounds of the feet of rice farmers are common during the planting and harvesting seasons and this is how this ubiguitous soil organism gains entry into the host (Chaowagul et al, 1989). Skin and soft tissue infections are also common manifestations of melioidosis and may be a source of systemic infection. Cutaneous infections with B. pseudomallei are usually indolent soft tissue infections manifesting as cellulitis or abscesses. These soft tissue infections can progress to necrotizing fasciitis in melioidosis (Wang et al, 2003).

A definitive diagnosis of melioidosis is made by isolation and identification of the bac-

terium Burkholderia pseudomallei from clinical specimens, which is the gold standard. A screening system, which includes a Gram stain, oxidase reaction, resistance to aminoglycosides and colonial characteristics on a differential agar medium (Ashdown) are simple and economical ways of identifying suspected strains of Burkholderia pseudomallei in areas of endemicity (Dance et al, 1989). Our laboratory adheres to this method. The screening has proven to be accurate and is supplemented by the Analytical Profile Index (API) 20NE for confirmation. Besides culture, many serodiagnostic methods have been described. These include the complement fixation test, the indirect hemagglutination test (IHA), the immunofluorescence test and an enzyme immunoabsorbent assay and test for exotoxin. IHA remains the most widely used test despite its poor sensitivity and specificity. Interpretation is hampered by high titers in normal individuals living in hyperendemic areas where rates of background seropositivity may vary from to 30 to 47% in various populations (Khupulsup and Petchclai, 1986). A new immunochromatographic test for the rapid detection of IgM and IgG antibodies to Burkholderia pseudomallei is said to have a sensitivity of 100% IgG and 93% for IgM, while the specificity is 95% for both these assays. The test is rapid and simple to perform, with results obtained in 10 minutes and should be useful as an alternative diagnostic mothod for melioidosis (Cuzzubbo et al, 2000).

A high index of suspicion is important in making the diagnosis of melioidosis, as a timely diagnosis and prompt accurate treatment are crucial in this potentially fatal infection. Despite appropriate antimicrobial therapy, severe or septicemic meliodosis has a mortality rate of 40-75%. In this patient, *Burkholderia pseudomallei* was initially not thought of as the causative agent for his illness but when alerted by the laboratory, appropriate therapy was instituted immediately. Ceftazidime either as monotherapy (120 mg/kg/day) or combined with trimethoprim-sulphamethoxazole are the recommended antibiotics in the acute phase in Malaysia (Suputtamongkol and Chaowagul, 1994; Hospital Kuala Lumpur, 2001). Although there are slight variations in terms of recommended regimens between countries the only treatment that appears to demonstrate a mortality benefit is ceftazidime. This was demonstrated in a sequential open-label randomized trial of ceftazidime versus chloramphenicoldoxycycline-trimethoprim-sulphamethoxazole in severe disease (White et al, 1989). In Thai adults, the use of ceftazidime has been associated with a 50% reduction in mortality from 74 to 37% (White et al, 1989). In order to prevent relapse of this persistent and potentially lethal infection, optimal maintenance or eradication therapy is imperative. In most reported series, the course of oral maintenance treatment is suggested to be between 12 and 20 weeks duration consisting of trimethoprimsulphamethoxazole (8-12/40-60 mg/kg/day) plus doxycycline (4 mg/kg/day) (Supputtamongkol, 1996; Chaowagul et al, 1999; Raja et al. 2005).

Cases of melioidosis continued to be seen in Malaysia and may have varied presentations. Its true incidence is probably underestimated as the disease is non-notifiable in Malaysia. It is important to be aware of soft tissue infections caused by *Burkholderia pseudomallei*, including the remote possibility of necrotizing fasciitis at the extreme end of the spectrum. Successful management requires a high index of suspicion, appropriate antimicrobial selection, and prompt and aggressive surgical debridement.

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