Short Communication

Melioidotic Necrotizing Fasciitis Presenting as a Supraclavicular Mass

Tsung-Liang Ma, Guan-Cheng Huang, Hung-Jen Tang and Chia-Ming Chang1,2*

Department of Internal Medicine, Chi Mei Medical Center, and 1Division of Infectious Diseases and 2Division of Geriatrics and Gerontology, Department of Internal Medicine, National Cheng Kung University Hospital, Tainan, Taiwan

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SUMMARY: Melioidosis is an infection caused by Burkholderia pseudomallei that usually involves the respiratory tract. It may manifest as pneumonia, septicemia, or localized infection. We present here a case of melioidosis initially manifesting as a mass over the supraclavicular area and subsequently progressing to necrotizing fasciitis. With appropriate antimicrobial treatment and adequate surgical debridement, localized melioidosis can be treated successfully. Melioidosis should be considered in the differential diagnosis of neck masses, especially in patients who have traveled to or stayed in an endemic area.

Melioidosis is an infectious disease caused by the Gram-negative bacilli Burkholderia pseudomallei. It is endemic in Southeast Asia, northern Australia, Central and South America, and China (1). Clinical manifestations are variable, and it has acute, sub-acute, chronic, and sub-clinical forms (1,2). In Taiwan, endemic cases of melioidosis have emerged in the past decade (2). From July through September 2005, 40 cases of melioidosis were reported in southern Taiwan after a typhoon. Most of these patients presented as bacteremia and pleuropulmonary infections (3). Diagnosis by clinical presentations only, without microbiological information, is difficult. The presentation usually involves the respiratory tract and may manifest as pneumonia, septicemia, or localized infection (1). Although soft tissue infections are not uncommon, necrotizing fasciitis caused by B. pseudomallei has rarely been reported (4,5). We present here a case of melioidosis manifesting initially as a supraclavicular mass that subsequently progressed to necrotizing fasciitis.

A 41-year-old man suffered from general malaise for about half a year with recent exacerbation. On June 19, 2006, he was referred to the hematology clinic due to leukocytosis found at a local clinic. He had had no chronic diseases such as diabetes mellitus or hypertension in the past. In the first two outpatient visits, only mild leukocytosis (leukocyte counts: 10,200/μL and 12,000/μL; neutrophils 70.8 and 80%, respectively) was found. No fever or lymphadenopathy was noted. Biochemical data were normal for glucose (86 mg/dL) and for renal and liver functions (blood urea nitrogen 14 mg/dL, creatinine 1.0 mg/dL, aspartate aminotransferase 18 IU/L, alanine aminotransferase 12 IU/L). Chest X-ray and abdominal ultrasonography were unremarkable, while esophago-gastroduodenoscopy showed only erythematous gastritis.

However, on the third outpatient visit to the hematology clinic (about 28 days after the first visit), a palpable mass was noted on his right supraclavicular region, which the patient noted about 4 days earlier. It began as a small painless hard nodule that enlarged gradually. He had no sore throat, cough, or other discomfort. Physical examination revealed a hard, non-tender mass about 4 × 5 cm in diameter on the right supraclavicular region. The liver and spleen were not palpable. Computed tomography of the head and neck showed a 3.7 × 3.3 cm ill-defined lesion with central necrosis over the right lower neck to the supraclavicular region with adjacent soft tissue infiltration (Figure 1). Multiple necrotic lymphadenopathies over the right carotid space of the neck and mediastinal regions were also noted (Figure 2).

Echo-guided fine-needle aspiration was arranged for the necrotic mass lesion. Ultrasonography disclosed multiple bilateral neck lymph nodes and a 7.9 cm multi-loculated mass with a heterogeneous hypo-echoic pattern in the right lower neck. The mass was aspirated, and a specimen was collected for cytology and bacterial culture. For further pathology...
examination, the patient was then referred to the general surgery clinic for incision biopsy of the mass. At that time, about a week after the mass was initially noted, it enlarged further and became painful. The biopsy specimen was composed of muscle and soft tissue but no nodal tissue. Pathology showed necrotizing inflammation without malignancy.

Bacterial culture of the aspirated tissue later yielded growth of *B. pseudomallei*, which was susceptible to cefazidime, cefpirome, imipenem, and piperacillin-tazobactam but was intermittently resistant to trimethoprim-sulfamethoxazole and ciprofloxacin. The patient was then admitted for further antimicrobial therapy.

On admission, purulent discharge from the previous incision wound of the supraclavicular mass was noted. Hemogram disclosed a hemoglobin level of 14.6 g/dl and a leukocyte count of 9,400/μL with 72.3% neutrophils and 14.9% lymphocytes. Other biochemical data, including liver and renal functions, were all within normal limits. No serological test was available at our hospital.

The personal history of this patient was reviewed in advance. He lives in the Che-Ting Country of Kaohsiung County, which is a seaside village in southern Taiwan. The patient also mentioned that an alcoholic man living in his neighborhood died of melioidosis. The patient was a cement worker in building construction and had the habit of cigarette smoking about more than one pack per day for 20 years. He also had the habit of social drinking and betel nut chewing for 10 years but had quit half a year earlier when he felt discomfort. He had a pet dog for years and had never traveled abroad.

During admission, after two sets of blood culture were collected, the patient was initially treated with cefazidime 2 g intravenously every 8 h for 9 days. Blood culture later yielded no growth of pathogen. However, purulent discharge still oozed from the incision wound. On the 10th hospital day, a decision to perform surgical excision of the mass and wound debridement was made. Intra-operatively, diffuse necrotic tissue was noted within the subcutaneous space, underlying muscle, and scattered over the surrounding tissue and fascia space. Loss of resistance of the normally adherent superficial fascia to blunt dissection was noted, consistent with necrotizing fasciitis. Fasciectomy and debridement were performed. The pathology of the debrided tissue showed necrotic tissue, collection of pus cells, and edematous tissue infiltrated by acute and chronic inflammatory cells. Several multinucleated giant cells were found in the necrotic focus. Acid-fast and Auramine-Rhodamine stains were negative for bacilli. Aerobic culture of the debrided tissues did not yield any bacterial growth.

Postoperatively, the antimicrobial agent was switched to imipenem 500 mg intravenously every 6 h for 3 weeks. After discharge, the patient received oral amoxicillin-clavulanate for additional 3 weeks. At the end of the antimicrobial treatment, the value of the erythrocyte sedimentation rate was 17 mm/h (normal, 0-15 mm/h). The patient was followed up for 8 months after discharge during which he remained asymptomatic.

Melioidosis has been called the great mimicker because of its variant clinical manifestations and courses. Diagnosis is difficult unless microbiological information is confirmed. As our case presented, lymphoma was the initial impression based on history and initial manifestation. Although diabetes mellitus, thalassemia, renal disease, chronic alcohol consumption, and steroid use are common underlying conditions of melioidosis (1), none was found in our patient.

Skin and soft tissue infections are common manifestations of melioidosis, but progression to necrotizing fasciitis has rarely been reported (4), especially involving the head and neck. Lim et al. reported 4 cases of melioidosis involving the head and neck region, with one case of parotid abscess that was drained but subsequently progressed to necrotizing fasciitis (5). Our patient had undergone needle aspiration and incision biopsy of the mass but finally underwent debridement due to poor wound condition. Whether or not the previous procedure on the lesion led to the progression of the disease to necrotizing fasciitis requires further investigation.

Suppurative lymphadenitis caused by melioidosis has rarely been encountered. In addition to the head and neck region, inguinal and mediastinal lymph nodes could also be infected (6). The imaging study in our case showed necrotic lymphadenopathies over the right carotid space of the neck and mediastinal regions. Melioidosis should be considered in the differential diagnosis of suppurative regional lymphadenitis, especially among patients who have traveled to, or stayed in, an endemic area (6).

The modes of acquisition of melioidosis include ingestion, inhalation, and inoculation (1). In the past, inhalation was thought to be the primary mode of infection, but now inoculation is believed to be the primary mode (1). Based on his occupational history of being a construction worker, the patient might have been infected through the inhalation of contaminated dusts or inoculation with contaminated soil or water through wounds or skin abrasions.

Because of intrinsic resistance of *B. pseudomallei* to commonly used first-line antimicrobial agents, only amoxicillin-clavulanate, cefazidime, meropenem, or imipenem, alone or in combination with co-trimoxazole, have been reported to have in vitro and in vivo bactericidal effects (7,8). The current recommended therapy includes two phases, an acute phase with intravenous cefazidime or carbapenem for at least 14 days, followed by an eradication phase with oral amoxicillin-clavulanate or a combination of co-trimoxazole and doxycycline for 12 to 20 weeks (1,9). Relapse can occur after a median of 6 to 8 months. An eradication therapy of less than 8 weeks is associated with a higher risk of relapse (1). Our patient received only a 3-week eradication therapy. Although he remained asymptomatic after cessation of the oral antimicrobial agent for 8 months, long-term follow-up is mandatory because of the potentially incomplete treatment and the fairly long period before relapse occurs.

As melioidosis becomes an emerging infectious disease, clinicians should consider it in the differential diagnosis of neck mass. It is possible to misidentify a supravacular soft tissue mass as simple lymphadenopathy. In addition to early recognition, appropriate antimicrobial treatment and surgical debridement are required to improve prognosis.

**REFERENCES**

