Clinical and Imaging Findings of Musculoskeletal Melioidosis in the Right Hip: A Case Report
우측 대퇴부에서 발생한 근골격계 유비저의 임상적, 영상학적 소견: 증례 보고

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Melioidosis is an infectious disease caused by a Gram-negative bacterium thought to be caused *Burkholderia pseudomallei*. This disease is endemic in tropical regions, but is not endemic and is rarely encountered in Korea. Nevertheless, the importance of early diagnosis of melioidosis is drawing substantial attention, due to its proliferation and the high mortality caused by the disease. Melioidosis can attack any organ, and manifests with a variety of symptoms. In particular, musculoskeletal melioidosis is rare and presents with nonspecific musculoskeletal symptoms. The imaging features of musculoskeletal melioidosis manifest a form of osteomyelitis or septic arthritis with soft tissue abscess or can mimic a bone tumor. This study describes the clinical and imaging findings of melioidosis involving the right femur and hip joint in a 64-year-old man.

Index terms
Melioidosis
*Burkholderia Pseudomallei*
Osteomyelitis
Femur
Infection

INTRODUCTION

Melioidosis is an infectious disease caused by *Burkholderia pseudomallei*. The disease is endemic in tropical regions, particularly in Southern Asia and Northern Australia, but is rare in Korea. Until 2014, only 11 melioidosis patients were reported by the Center for Disease Control and Prevention (CDC) in Korea and all patients had a history of travel to Southeast Asia (1). Melioidosis can affect any organ and cause various clinical manifestations. The imaging findings of musculoskeletal melioidosis manifest as osteomyelitis or septic arthritis with soft tissue abscess or can mimic bone tumor (2, 3). The current study describes the clinical and imaging findings of melioidosis involving the right femur and hip joint in a 64-year-old man.

CASE REPORT

A 64-year-old male presented with complaint of right hip and inguinal pain. He reported a history of diabetes mellitus of fifteen years duration, as well as a history of esophageal cancer. The esophageal cancer was in complete remission after radiochemotherapy about eight years ago. Until recently, he had resided in Cambodia for five years.

About one month ago, the patient was admitted to the hospital with pneumonia and fever as the only presenting symptom. Pneumonia was diagnosed on chest X-ray and computed tomography was performed to identify the cause of the fever. About three weeks after hospital discharge, he complained of right hip and inguinal pain that had suddenly and markedly increased in the absence of any history of identifiable trauma or...
Fig. 1. A 64-year-old melioidosis patient with right hip pain. 
A. Initial and 3 weeks follow-up radiography images including the right hip show no abnormal findings on soft tissue or bony structure. 
B. Whole body bone scan shows multifocal increased uptake in the greater and lesser trochanter of the right femur, and weak uptake in the femoral head.
arduous physical labor. He reported that he had experienced pain for several months, but assumed it was "osteoarthritis caused by aging" because it was a nominal pain similar to that in his other joints. On physical examination he had mild tenderness on the anterior thigh just below the inguinal region. The range of motion in his right hip was normal, but the log rolling test was positive, suggesting inflammation on the right femoral head or acetabulum.

On initial radiographic examination of the right hip (Fig. 1A), no abnormal findings in the soft tissue and bony structures were appreciated; the study revealed evidence of mild degenerative changes only, which was also observed three weeks later on follow-up radiography. However, the pain had progressively increased and became more intense with time. After only one
week, the patient complained that he was unable to walk due to severe pain in the right hip and inguinal area.

Further evaluation was felt to be necessary, and a whole body bone scan and contrast enhanced magnetic resonance imaging (MRI), were performed. The whole body bone scan (Fig. 1B) revealed multifocal increased uptake in the greater and lesser trochanter of the right femur, and weak uptake in the femoral head. On MRI (Fig. 1C, D), the fat-suppressed T2-weighted MRI showed heterogeneously high signal intensity and T1-weighted MRI and proton density-weighted MRI showed heterogeneously low signal intensity in the inferior portion of the femoral head and neck, greater and lesser trochanter, and proximal shaft of the femur, with contrast enhancement observed in some areas at the same sites.

In other words, there were some areas that showed high signal intensity on T2-weighted MRI but no enhancement.

These intraosseous non-enhanced areas were thought to consist of necrotic tissue. Contrast enhancement was evident not only in the femur, but also in the surrounding soft tissue. In the right hip joint, prominent synovial thickening and enhancement with increased joint effusion were found. A distended iliopsoas bursa (with bursal wall thickening and increased effu-
sion) suggested an iliopsoas bursitis or secondary change of the inflammation of the femur and hip joint. These MRI findings were thought to be consistent with acute osteomyelitis with bone necrosis.

An orthopedic doctor at our hospital evaluated the lesion under the impression of disease, not only inferred disease from the

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**Fig. 1.** A 64-year-old melioidosis patient with right hip pain.
**E.** Positron emission tomography/computed tomography shows multifocal hypermetabolic lesion on the proximal shaft of the femur, femur neck, femoral head and iliopsoas bursa.
**F.** Post-excisional biopsy radiography (left) shows fracture of the lesser trochanter of the right femur and an irregular shaped radiolucent area on the right proximal shaft of the femur. This is thought to be due to the excisional biopsy. Post-re-operative radiography (right) shows widened radiolucent area on the proximal shaft of the femur where the drainage catheter tip is located. This indicates that an abscess is formed in this radiolucent lesion.
imaging findings but also metastatic bone lesion, because the patient had a history of esophageal cancer. As the next step of evaluation, positron emission tomography/computed tomography (PET/CT) was performed to rule out malignancy. On PET/CT (Fig. 1E) a multifocal, hypermetabolic lesion was identified on the proximal shaft of the femur, femur neck, femoral head and iliopsoas bursa.

About one month later, an excisional biopsy of the right femur lesion was performed (Fig. 1F, right). The orthopaedic surgeon at our hospital ordered the biopsy because it was thought that the probability of metastasis was most likely. Subsequent pathology confirmed the right femoral lesion as a condition involving chronic inflammation with necrosis and degenerative bony trabeculae of woven bone without osteoblastic rimming. The results also confirmed the soft tissue surrounding the femoral head as a suppurative inflammation with granulation tissue and fat necrosis. Other evaluations such as tissue culture were not performed. About one month later (and after the excisional biopsy), surgical re-intervention was performed due to some oozing at the operation site (Fig. 1F, Left). A trochanteric abscess was found on the surgical bed, therefore, incision, drainage and a biopsy of the synovium of the hip were performed. The acute suppurative inflammation in the synovium of the hip was pathologically confirmed. A culture of the abscess contents revealed *Burkholderia pseudomallei*. This Gram-negative bacillus was previously detected in blood cultures to either rule in, or exclude, the presence of pneumonia.

After the initial diagnosis of pneumonia, the patient was treated with ceftriaxone (a type of β-lactam antibiotic and clarithromycin (a type of macrolide), as the first line of defense with regard to treatment for community-acquired pneumonia. After the culture test results were obtained, the patient was treated with piperacillin/tazobactam, another β-lactam antibiotic, and levofloxacin, a fluoroquinolone, for about three weeks (as a guideline of severe pneumonia caused by Gram-negative bacilli). After culturing of *Burkholderia pseudomallei* in the right hip abscess, the same antibiotics therapies were planned, because the same type of antibiotic is used for melioidosis regardless of the location or type of infections.

However, the patient was transferred to another hospital closer to his residential area.

**DISCUSSION**

Melioidosis is a disease endemic to tropical parts of the world in general, and Southeast Asia and Northern Australia in particular. It is thought that the etiology of melioidosis is most likely, *Burkholderia pseudomallei* (4). The first patient diagnosed with melioidosis resided in Myahjar, and the case was reported in 1912 In Korea, the first documented melioidosis patient was reported in 2003 (5).

This disease is regarded, and managed, as a contagious communicable disease in Korea, due to its increasing incidence with increased international travel and a high mortality rate, of about 80%, without proper early medical intervention and treatment (6).

To date, a total of eleven melioidosis patients have been reported in Korea. These patients were all male, with a mean age of 52.3 years (range, 32–66 years) and had all visited Southeast Asia. The period of symptom development was variable, ranging from one to three days (1). Underlying diseases, including diabetes mellitus, chronic lung disease, renal disease, malignancy, immunosuppressive treatment and heavy alcohol consumption and/or or chronologic age over 50 years, are well-known risk factors. These risk factors are associated not only with susceptibility to melioidosis, but also with the relative severity of the disease (1, 7).

The patient in this report had much in common with other male Korean patients. The patient was 63-years-old man, and with documented histories of residence in Southeast Asia, diagnosed diabetes mellitus and cancer treatment.

Melioidosis can affect any organ and cause a variety of different symptoms. Pulmonary involvement, such as pneumonia, is the most common clinical feature and disseminated abscess formation is a characteristic feature of melioidosis (1). According to Cheng and Currie (4), musculoskeletal melioidosis is difficult to differentiate from other infectious musculoskeletal diseases based on clinical symptoms, but the systemic features are prominent.

Musculoskeletal melioidosis has a relatively low incidence. Currie et al. (8) reported that only 4% of melioidosis patients presented osteomyelitis or septic arthritis in Northern Australia, over a period of ten years. Another study (9), carried out in Northern Australia, reported a 7.6% incidence of bone and joint infections in melioidosis patients.
There are two possible infection routes for musculoskeletal melioidosis. One is directly from skin injury or wound infection. The other is hematogenously from another primary disease, such as pneumonia with a form of septicemia (9). The patient in this case report can be considered to belong to the latter group.

In this report, we describe a patient with melioidosis that involved the right femur and hip joint, with recent underlying pneumonia. *Burkholderia pseudomallei* were cultured from a trochanteric abscess on the right hip joint and previous blood drawn during pneumonia, suggesting hematogenous spread from the lung, presenting as pneumonia. The patient also had a history of diabetes mellitus, radiochemotherapy for esophageal cancer and had lived in Cambodia until recently. On MRI, the inferior portion of the femoral head and neck, the greater and lesser trochanter, and the proximal shaft of the femur showed heterogeneous high signal intensity on T2-weighted MRI and low signal intensity on T1-weighted MRI. On post-contrast MRI, some areas of the right femur that showed high signal intensity on T2-weighted MRI did not show enhancement, suggesting that they contained necrotic tissue. These non-enhanced areas, which were thought to consist of necrotic tissue, can provide strong evidence of melioidosis on imaging evaluation.

In conclusion, melioidosis should be suspected when there is an infectious disease such as pneumonia in patients with chronic diseases including diabetes mellitus who have visited an area where the disease is prevalent including Southeast Asia and northern Australia. Furthermore, on initial or follow-up evaluation, necrosis or abscess may be strong evidence of melioidosis.

REFERENCES

우측 대퇴부에서 발생한 근골격계 유비저의 임상적, 영상학적 소견: 증례 보고

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유비저는 Burkholderia pseudomallei에 의해 발생하는 감염성 질환이다. 이는 주로 열대 지방에서 발생하는 풍토병으로 한
국에서는 드물다. 하지만 최근 발병률이 점점 증가하고 있고 높은 사망률을 보여 초기 진단의 중요성이 부각되고 있다. 유
비저는 인체의 어떤 장기에서도 발병 가능하며 다양한 증상을 일으킬 수 있다. 특히 근골격계 유비저 환자는 빈도가 매우
드물고 비특이적인 근골격계 증상을 일으킬 수 있다. 현재 알려져 있는 근골격계 유비저의 영상학적 소견은 골수염의 형태
나 농양을 동반한 화농성 관절염 또는 골종양을 모방하는 것으로 알려져 있다. 제4 64세 남자 
환자의 우측 대퇴골과 우측 고관절에서 발생한 유비저의 임상학적 그리고 영상학적인 소견에 대해 다루고자 한다.

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