Case Report / *Olgu Sunumu*

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Pathological Fracture of Distal Femur in a Child: What is the Culprit?

Çocukta Patolojik Distal Femur Kırığı: Suçlu Kim?

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Abstract

We reported a case of a six-year-old girl with initial radiograph finding of pathological fracture of the distal femur at the metaphysis region. Initially, malignancy was suspected. However, blood and bone cultures revealed *Staphylococcus aureus*. Thus, a diagnosis of osteomyelitis was made. She was put on a right hip spica cast for one month and responded well to antibiotics. This case report aimed to discuss the various imaging modalities in establishing the early diagnosis of a child with pathological fracture due to osteomyelitis.

Keywords: Osteomyelitis, pathological fracture, *Staphylococcus aureus*

Öz

Bu olgumuzda, radyografisinde distal femur metafiz bölgesinde distal femurda patolojik kırık bulgusu olan ve malignite şüphesi olan altı yaşında bir kız hastayı sunduk. Olgumuz, yapılan kan ve kemik iliği kültürü sonucunda *Staphylococcus aureus* üremesi sonucu tanı osteomyelit ile uyumlu değerlendirildi. Hastamız, bir ay süreyle uygulanan sağ kalça alçısı ve antibiyotik tedavisine iyi yanıt verdi. Bu olgu sunumu, osteomiyelite bağlı patolojik kırığın erken tanısında çeşitli görüntüleme yöntemlerinin rolünü tartışmayı amaçlamaktadır.

Anahtar Kelimeler: Osteomiyelit, patolojik kırık, *Staphylococcus aureus*

Introduction

Osteomyelitis is a common pediatric musculoskeletal infection. Acute hematogenous osteomyelitis is an infection that usually affects the growing skeleton and particularly the bone's most vascularized sections (1). If the symptoms have been present for less than two weeks, the condition is regarded acute (2). Acute osteomyelitis has an incidence of 8-10 per 100.000 in developed countries and an even higher incidence, up to 80 per 100.000, in developing countries (3).

This infection can weaken normal bone structure, resulting in the risk of a pathologic fracture. The metaphysis is the primary site of infection in children owing to its abundant vascularity; a metaphyseal equivalent, at the junction of bone and cartilage in a skeletally immature flat or round bone, and has similar vascularity to the metaphysis of a long bone, and therefore, is also particularly susceptible to osteomyelitis (1). Pathologic fractures associated with acute bacterial hematogenous osteomyelitis are rare in all age groups. Most of the cases that have been reported are in the adult population, with only six case reports found in the pediatric age group (4). Its presence is usually suspected based on suggestive clinical history and physical examination, elevated inflammatory markers, and radiographic findings. Magnetic resonance imaging (MRI) can confirm the diagnosis and determine the extent of bone and soft-tissue involvement.

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Staphylococcus aureus is by far the most common causative agent in osteomyelitis and has been reported to be responsible for 67% to 89% of cases of acute hematogenous osteomyelitis in the pediatric population (4).

Case Report

A six-year-old Malay girl presented with pain and swelling of the distal right thigh. About six weeks prior to her presentation to the emergency department, she had a history of non-trivial falls while playing at the side of her house. She was afebrile during that time and avoided movement of the right lower limb. The right thigh appeared swollen and distorted, with normal skin color and temperature on examination. Mobilization of the right lower limb provoked tenderness.

Initial laboratory investigation of full blood count (FBC) showed total white blood cell count as 11.4×10^9/L. Her enterocyte sedimentation rate and C-reactive protein (CRP) level were high, measuring 40 mm/hr and 44 milligrams per liter (mg/L), respectively.

A radiograph of the right femur was performed and showed a pathological fracture at the distal thigh at the metaphysis region (Figure 1). There was a permeative lytic bone lesion at the meta-diaphysis of the right distal femur. Minimal lamella periosteum elevation at the proximal femur with minimal surrounding soft tissue swelling was noted on the radiograph.

Figure 1. Radiograph of the right femur (AP) shows a displaced fracture of the distal thigh at the metaphysis region.

An MRI examination of the right femur was performed and showed abnormal signal intensity of the marrow from proximal right femoral diaphysis extending down to the metadiaphysis region, which appeared heterogeneous isointense on T1WI, heterogeneous hyperintense on T2WI, and not suppressed on STIR sequence (Figure 2). It showed heterogeneous enhance-

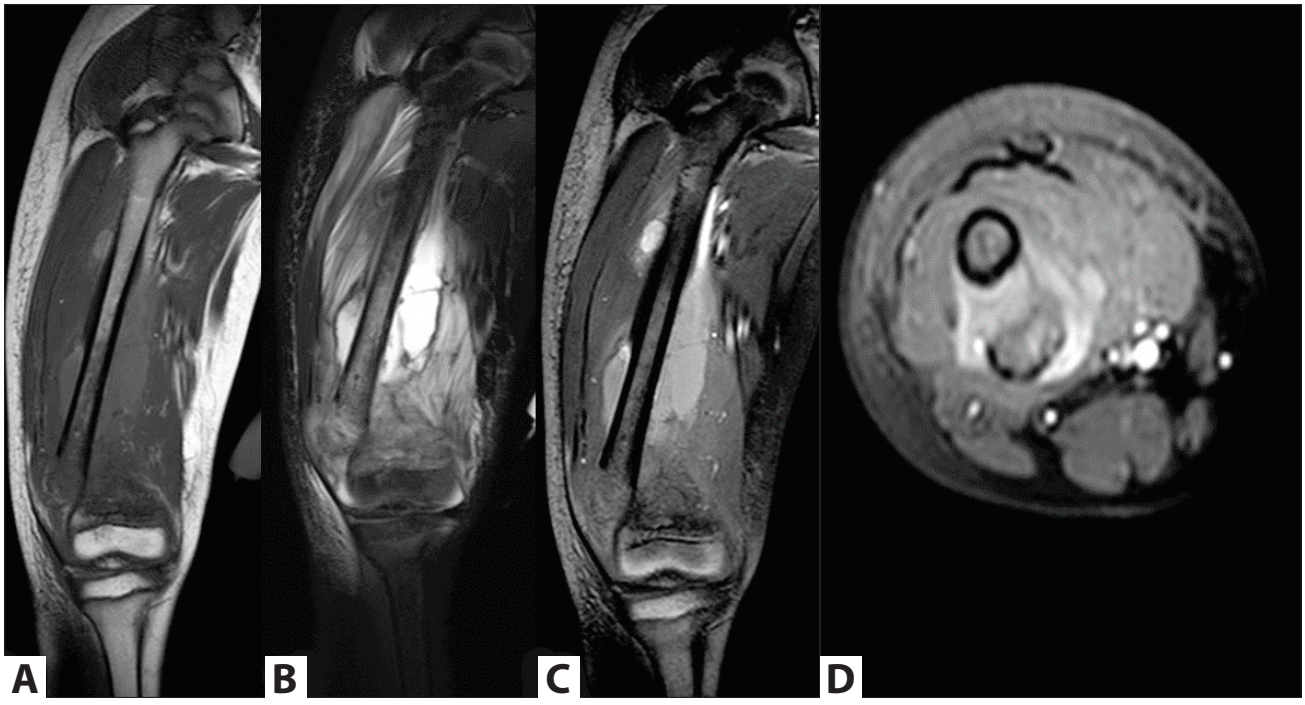

Figure 2. MRI of the right femur shows the abnormal signal intensity of the marrow, which appears heterogeneous isointense [coronal T1WI **(A)**], and heterogeneous hyperintense [coronal T2WI **(B)**]. It shows heterogeneous enhancement in post-contrast [coronal T1 and axial T1 post gadolinium **(C, D)**].

ment in post-contrast. However, no obvious extension into the distal femoral epiphysis was seen. No extra-osseous soft tissue component was noted. Posteromedial displacement of the distal third of the right femur was seen in the radiograph. Surrounding cystic lesion/collection appeared isointense on T1WI, hyperintense on T2WI, presence of fluid-fluid level, and blooming artifact suggestive of hematoma were recorded. Edematous and streakiness of the surrounding muscles of the thigh at the medial and lateral compartments was associative of inflammatory changes. No rim-enhancing lesion, cloaca, and involucrum of the sequestrum were seen.

Together with clinical parameters, plain radiograph, and MR imaging; these findings were concluded as an aggressive right femur intramedullary lesion with pathological fracture. The differential diagnosis was given as osteosarcoma or Ewing sarcoma.

Because of suspicion of a malignant process, an incisional biopsy of the fracture site was performed. Samples were sent for culture and sensitivity and turned out to be positive for gram-positive cocci *Staphylococcus aureus*. The patient underwent surgery for right hip spica application. A biopsy of multiple pieces of bony fragments measuring 20 mm in aggregates in diameter was submitted to the histopathology laboratory. Histopathological examination revealed the presence of reactive new bone formation, and bone necrosis (sequestrum) with neutrophilic exudates in favor of acute osteomyelitis changes (Figure 3).

The patient was put in a right hip spica cast for one-month duration. A course of antibiotics given was intravenous cloxacillin 425 mg QID for three weeks in the ward. The patient was discharged well, followed by a tablet of cloxacillin 250 mg QID for one week.

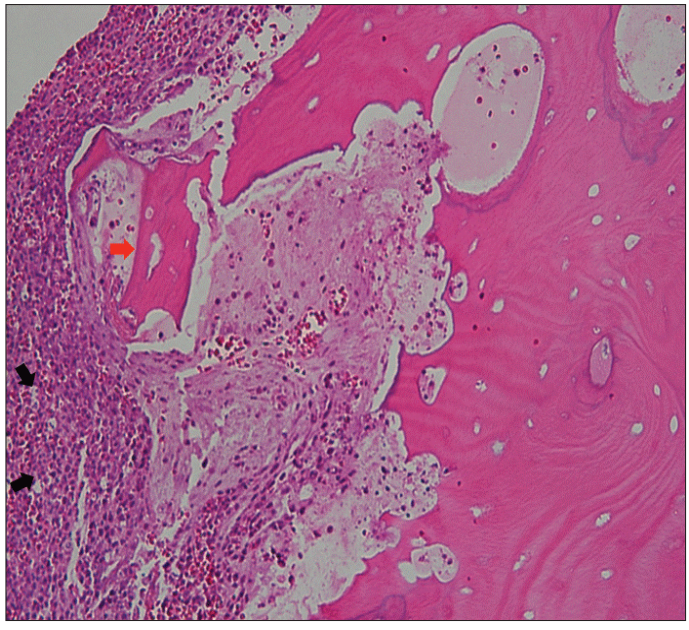

Figure 3. Acute osteomyelitis with the presence of sequestrum (bone necrosis); red arrow, new bone formation and neutrophilic abscess (black arrow) x200.

Discussion

Osteomyelitis is inflammation of the bone that is due to an infection, which is typically bacterial. Bone and joint infections are associated with high rates of morbidity and occasional mortality. The usual mode of infection is hematogenous, via the arterial blood supply. Acute hematogenous osteomyelitis is the most common form of bone infection in children. About half of all instances occur in children under the age of five and it is twice as common in boys as in girls. Infections are commonest in long bones such as the femur, tibia, or humerus.

The common clinical presentation of acute osteomyelitis in children includes fever, pain, swelling, and erythema with a history of trauma in the affected area. The most typical symptoms are a limping gait, limited movement of the affected lower extremities, or an unwillingness to bear weight (5). In children, the pathological fracture is a rare first manifestation of acute osteomyelitis. However, *Staphylococcus* infection that leads to acute osteomyelitis in children can complicate with pathological fractures (4).

The final diagnosis, in this case, was pathological fracture secondary to bacterial osteomyelitis. A closed fracture with a secondary infection is very rare. It is unusual for the infection to complicate a closed fracture in a healthy and immunocompetent person. Indeed, there are only a very few case reports of infection complicated by a closed fracture in the medical literature (6).

Clinical examination and laboratory investigations are crucial in early evaluation before imaging evaluation is performed. However, as in this case, the pathological fracture of the distal right femur as initial presentation gives rise to suspicion of the possibility of bone malignancy. In the setting of pathological fractures, apart from osteomyelitis, the differential diagnosis includes primary malignant bone tumors such as osteosarcoma, Ewing sarcoma, or any other benign bone tumor that is locally aggressive such as an aneurysmal bone cyst or giant cell tumor (7). Radiological imaging is important to narrow down the differential diagnosis.

Radiograph findings for osteomyelitis are periosteal reaction, new bone formation, and occasionally lucent lesions (Brodie's abscess) in the metaphyseal region; which is in subacute osteomyelitis. On the other hand, for chronic osteomyelitis, we see bone sclerosis, destruction, and periosteal new bone formation (8). Other imaging investigations, ultrasound, CT, MRI, or scintigraphy studies also play a role in diagnosing osteomyelitis.

In the early diagnosis of osteomyelitis, ultrasound has played a role in detecting periosteal changes, subperiosteal abscesses, and joint effusions; however, the overall sensitivity for the detection of osteomyelitis by ultrasound is low due to its high operator-dependent (6). On the other hand; its lack of radiation, speed, and low cost make this imaging modality very useful in children.

MRI is considered the golden standard for diagnosis of osteomyelitis as it has high sensitivity and specificity, which should be followed by bone biopsy for culture to guide optimal antibiotic treatment as well as for histological analysis to rule out other diseases (9). As in this case, initial MRI findings showed an aggressive right femur intramedullary lesion with pathological fracture, which gave differential diagnosis of malignancy and we proceeded with a biopsy. Furthermore, MRI helps distinguish the causes of a pathological fracture in the early stages and can help reduce the morbidity associated with a delayed diagnosis (6). It also delineates the extent of the infection.

Computed tomography is less useful in the pediatric population due to its radiation dose and lack of specific advantages, hence MRI is preferred (8). Bone scintigraphy (Tc-99m) is useful in the early stage of the disease. It will demonstrate increased radioactivity in the affected bone and for multifocal disease. Increased activity may be seen in dynamic perfusion, early blood pool, and delayed images. However, such uptake may be seen in other conditions including trauma and tumors; thus it is non-specific (8).

Conclusion

In conclusion, the diagnosis of osteomyelitis for a closed pathological fracture in the childhood is challenging. Only a few cases have been reported in the medical literature, and it illustrates the need to consider a broad differential diagnosis and obtain biopsy and cultures in treating a pathologic fracture in the pediatric age group. Imaging plays a vital role in the diagnosis of childhood osteomyelitis, and multidisciplinary team approaches are important for the better prognostic value of the patients.

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