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Case Series

Primary Subacute Osteomyelitis Affecting the Epiphysis in Children

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Abstract

Primary subacute osteomyelitis affecting the long bones in children is rare. Subacute osteomyelitis has obscure clinical findings, and an accurate diagnosis requires appropriate radiological examination.Herein, we present the cases of four children aged 4-8 years old. The distal humeral epiphysis was affected in two children, the distal femoral epiphysis was affected in one child, and the distal fibular epiphysis was affected in one other child. The symptoms included generalized illness with pain and restricted movement of the affected joint. Laboratory examinations were not specific. Radiological investigations, including radiography, magnetic resonance imaging (MRI), computed tomography (CT), were also performed. MRI was the most helpful diagnostic tool. All patients were surgically treated with debridement of the lesion and thorough cleaning of the joint. Pathological examination confirmed the diagnosis of subacute osteomyelitis. In three of these cases, the bacterial cultures were negative. Antibiotics were administered for 3 weeks. The patients had an uneventful recovery.We wanted to highlight the rarity of primary subacute osteomyelitis affecting the epiphysis. The diagnosis of an osteolytic lesion in the epiphysis requires detailed clinical and radiological examinations. Surgical treatment confirmed the diagnosis along with the pathological examination of the specimen.

Keywords: Subacute osteomyelitis, Epiphysis, Children, Surgical treatment.

Introduction

Diagnosis and treatment of bone infections in children are challenging. Although the incidence of bone infections has decreased in recent years, their peculiar presentations, both clinically and radiographically, continue to pose problems for accurate diagnosis and appropriate treatment.

Subacute osteomyelitis is characterized by mild symptoms, which last for 2-3 weeks, with the absence of initial acute symptoms. The patient is usually afebrile, with reduced mobility of the affected joint, unclear laboratory markers for infection, and a generalized feeling of weakness. The patients have negative blood cultures [1-3].

Subacute osteomyelitis most commonly affects the metaphysis, followed by the diaphysis, and rarely the epiphysis. Roberts et al. described six types of radiological subacute osteomyelitis. Type V epiphyseal subacute osteomyelitis is an eccentric osteolytic lesion of the epiphysis [4].

Acute osteomyelitis commonly affects the metaphysis in children dueto the vascular barrier of the growth plate. Epiphyseal involvement is usually secondary to metaphyseal involvement, through the joint space or the open transphyseal canals, in infants [5].Primary subacute epiphyseal involvement is an isolated infection without metaphyseal lesions and is rare.We present the clinical findings, radiological investigation, and treatment of four children diagnosed with subacute primary osteomyelitis affecting the epiphysis.

Patients and Methods

During the past 15 years, four children were diagnosed with subacute epiphyseal osteomyelitis.Their age ranged from 4 to 8 years old at the initial presentation. The elbow was affected in two children, and the knee and ankle joints were affected in two other children. In all children, there was a gradual onset of discomfort and vague pain in the affected limb. Limping was the main symptom in patients with lower limb involvement. Reduced mobility of the upper limb with discomfort was a symptom associated with elbow lesions.

Only one child with distal femoral epiphyseal involvement presented with a fever of 37.2°C at the initial examination. None of the patients had previously received antibiotic treatment. Since the onset of symptoms, the time interval for referral ranged from 3-5 weeks. A diagnosis of juvenile arthritis was initially proposed for the two children whose knee and ankle joints were affected. One of the children with an elbow lesion participated in taekwodo, and the symptoms were assumed to be associated with a traumatic injury.

Detailed clinical examination revealed reduced mobility of the affected joint in all children. The children were unable to jump on the affected limb. Those whose elbows were affected expressed difficulties in wearing clothes and toileting. For all children, the parents described the feeling of reduced daily activities and unwillingness to attend school.

The affected joint in all children had mild effusion and tenderness on palpation, and erythema was observed in the girl with the elbow epiphyseal lesion. Enlarged lymph nodes were not observed.

Radiological examination with conventionalradiography revealed the presence of an osteolytic lesion that was diagnosed after a detailed examination. The lesion in the distal femoral epiphysis was an eccentric osteolytic lesion with clear boundaries, normal bone cortices, and absenceof periosteal reaction. In the ankle joint, there was osteolytic destruction of the epiphysis and near metaphysis at the distal part of the fibula. An osteolytic lesion of the distal epiphysis of the humerus was observed on the elbow joint.

All our patients underwent blood tests, including measurements of hemoglobin, hematocrit, white cell count, platelets, serum C-reactive protein (CRP), erythrocyte sedimentation rate (ESR), blood cultures, and antinuclear antibodies.

Blood test results were unspecified. The 5-year-old child with knee effusion had an increase in the number of lymphocytes. The ESR ranged from 15-24 and CRP was normal in all four children.

Magnetic resonance imaging (MRI) was performed in all children within 24 h after the initial radiological examination.

MRI revealed an eccentric osteolytic lesion with clear borders and intact growth plate. The lesion appeared to be localized in the distal femoral epiphysis of the affected knee. The lesion extended to the metaphysis in the distal part of the fibula in the child whose ankle joint was affected. In the elbow joint, the lesion affected the epiphysis, and effusion of the joint was observed. There were no periosteal reactions or moating lesions in the cortices. The articular cartilage remained intact. Contrast-enhanced examinations were not performed. No anesthetics were administered for the examination.

The child with the elbow lesion was examined using CT, which revealed a lytic lesion of the epiphysis, extending partially in the metaphysis, with clear sclerotic borders, with disruption of the cortex. A bone scan was performed in two children to rule out multiple sites of osteomyelitis. The results showed a positive uptake only in the affected area.

All children underwent chest radiography, which revealed normal findings.

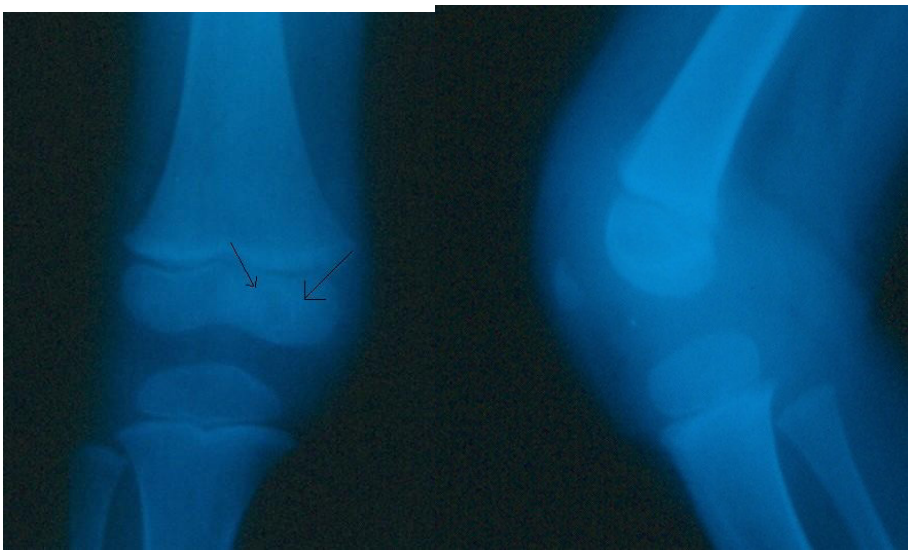

Figure 1A and 1B: X-ray AP and lateral, with an obscure eccentric lytic lesion in the distal femoral epiphysis,

Figure 1C: MRI with a T1 Fat sat image, with a lesion of the right distal femoral epiphysis, compared to the normal left side.

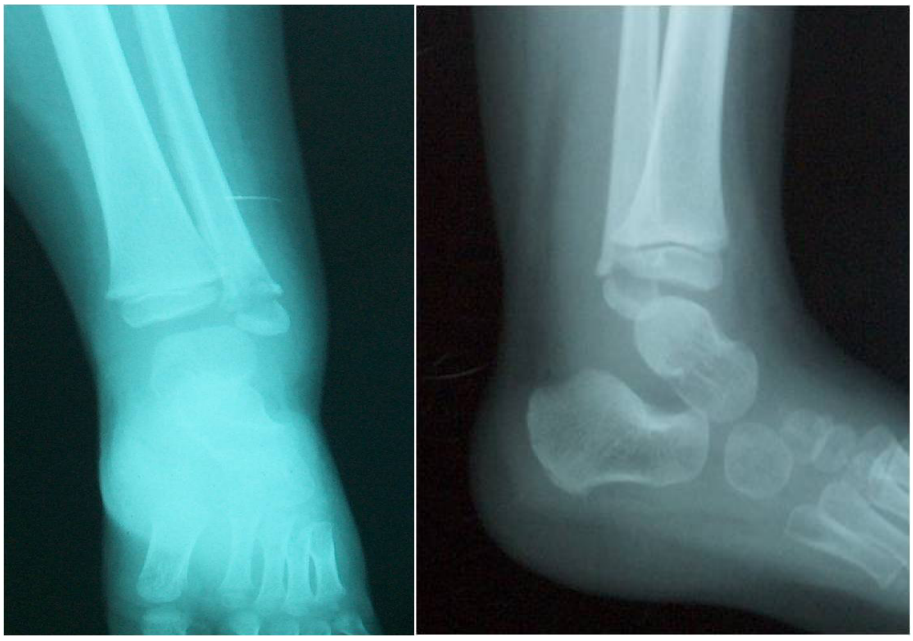

Figure 2A,B: X- ray AP and lateral of the ankle joint, with a lytic lesion of the distal fibular epiphysis, with soft tissue swelling of the ankle joint.

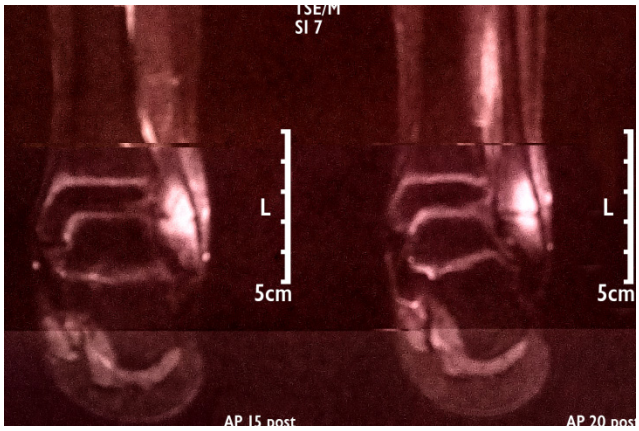

Figure 2C: MRI with a STIR image with oedema of the epiphysis and metaphysis, with an intact growth plate.

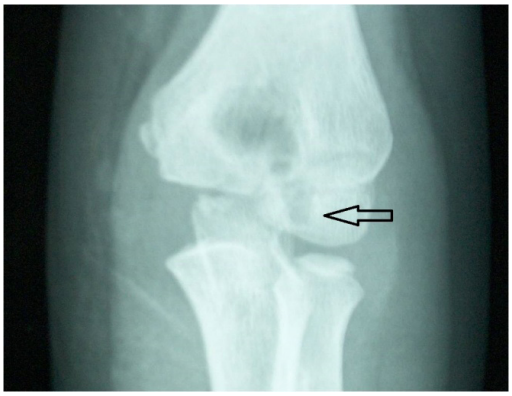

Figure 3A: X- ray of the elbow joint with an osteolytic lesion of the lateral humeral condyle.

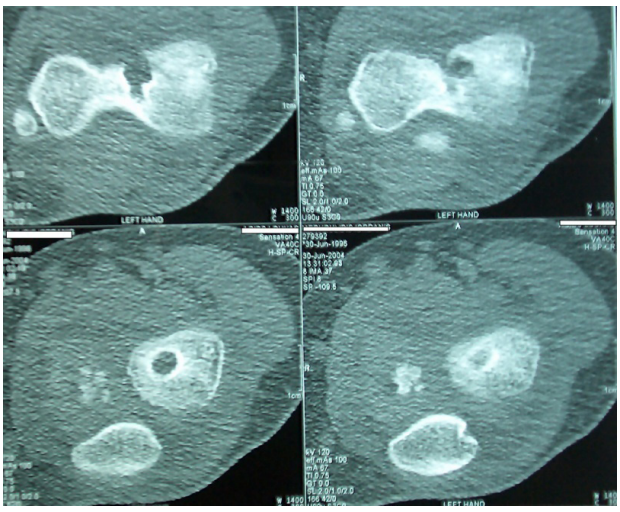

Figure 3B: CT scan with the lytic lesion of the lateral condyle, with cortex destruction.

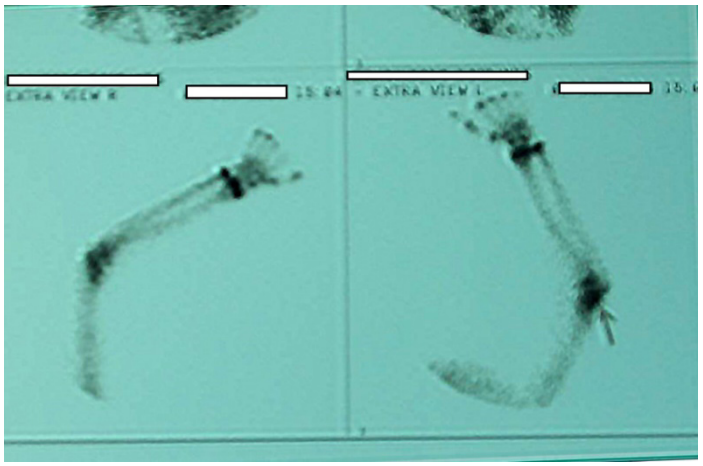

Figure 3C: Bone scan with positive uptake of the affected humeral epiphysis.

Results

All children were surgically treated within the day following the MRI examination. Surgery was performed using an open procedure with radiological guidance. Particular attention was paid to avoid iatrogenic lesionson the growth plate. The affected area was easily identified, and thorough cleaning was performed after collecting samples for pathology and culture. The osseous cavity was curetted, avoiding the growth plate. The articular cartilage is intact. Vancomycin was administered immediately after the procedure.

Cultures obtained from the surgery were negative in three patients. A girl with a lesion in the elbow tested positive for *Staphylococcus aureus*. Pathological specimens confirmed the diagnosis of osteomyelitis in all children. There were infiltrations with an increased number of neutrophils and lymphocytes, without atypia or bone edema.

All children recovered uneventfully. The affected joint was protected for 3 weeks, permitting gradual movement, and partial weight-bearing was started after a month for the children whose limbs were affected. Antibiotics were administered for 3 weeks. Clinical improvement was evident immediately following the procedure. The children were relieved of the painandwere getting better in general.

Joint movements returned to normal, and the children resumed their daily activities within 2 months. Radiological examination using conventional radiography was performed at 3, 6, and 12 months after the procedure. The growth plates were normal in appearance.

The patients were followed up for 8 years, with a normal axis of the lower limb and absence of leg length discrepancy. Children

with elbow lesions had a normal carrying angle with a full range of movements.

Figure 4A,B : Final AP and lateral x-ray of the patient with the distal fibular subacute osteomyelitis, with enlarged epiphysis and normal growth plate.

Discussion

The incidence of bone infections is decreasing because of appropriate early diagnosis and antibiotic treatment. Although the incidence is decreasing, there are challenges in establishing an accurate diagnosis since the symptoms are often obscure [6,7].

This is important when dealing with subacute osteomyelitis, where the symptoms are vague. Pain and joint movement restriction were the most common symptoms.

Subacute epiphyseal osteomyelitis was first described in two patients by King and Mayo in 1969 [8]. In 2001, Rasool published a case series of 21 children with subacute osteomyelitis as a primary lesion from 1990 to 1998. The patients had not been previously treated with antibiotics. The tibia was affected in 17 children, with epiphyseal involvement in two children. [9] Later, Hepfing reported three children with epiphyseal involvement, affecting the knee joint in two children and the upper epiphysis of the femur in one other child [10].

Laboratory investigations of subacute osteomyelitis are unclear. In their series, Ezra and Hamdy reported an increased ESR and a smaller increase in CRP measurements. In our patients, the blood test results were almost normal, with only a mild increase in the ESR, whereas the CRP measurements were normal [11, 12].

Radiographic examination is important; however, it does not provide a definitive diagnosis. An osteolytic lesion of the epiphysis may escape the initial diagnosis, as it appears later during osteomyelitis, with less striking findings.

MRI can accurately diagnose subacute osteomyelitis. It is easily available for use in tertiary hospital centers. It may require sedation when applied in young children; however, in general, children older than 5 years can be easily examined using MRI.

El Ali et al.described the pathological and radiological correlations of solitary long-bone epiphyseal lesions in children. Their case series of 49 children had 92% benign lesions, and only three children were diagnosed with malignant lymphoma and chondrosarcoma. Benign lesions included chondroblastoma (45%), osteomyelitis (18%), enchondroma, histiocytosis, and bone cysts. Chondroblastomas are found in older children and are more peripherally located. Osteomyelitis and chondroblastoma have similar clinical and imaging features. On radiography, chondroblastomas may appear with a more calcified internal matrix than osteolytic lesions, as in osteomyelitis. On MRI examination, they have characteristics similar to that of T2-weighted hyperintense and mild perilesional edema. They have similar post-contrast enhancements [13].

A bone scan is helpful when we are unable to localize the pathology or when multifocal involvement is suspected [14].

Diagnosis of subacute osteomyelitis remains a challenge when dealing with a chronically suffering child with an unclear history, who is afebrile or has a mild temperature, with non-specific laboratory findings. Thorough clinical examination revealing joint involvement with reduced mobility of the affected limb is the first step for establishing a correct diagnosis. While conventional radiography is helpful, MRI is the most accurate radiological investigation for diagnosis.

The primary therapeutic procedure for subacute osteomyelitis is surgical treatment. It provides a efinitive diagnosis of infection, distinguishing it from benign or malignant lesions. This is important because of the radiographic similarities between osteomyelitis and chondroblastoma. Osteolytic lesions, mainly in the metaphysis or diaphysis of the long bones in children, are important for distinguishing between osteosarcoma and Ewing sarcoma. Zairi et al. reported subacute osteomyelitis of the tibial diaphysis associated with a Brodie abscess, and the diagnosis was confirmed after bone biopsy in a 4-year-old boy. [15] Hoffman et al. (1990) reported that six children with subacute osteomyelitis in the diaphysis could not be distinguished based only on radiological findings. Surgery and pathological assessment confirmed the diagnosis of osteomyelitis [16].

Shah et al. reported 18 patients with primary epiphyseal osteomyelitis over 11 years. The distal femoral epiphysis was the most involved site. Eleven patients had *Mycobacterium tuberculosis* infections. They also reported a delay in diagnosis, mainly in those with tuberculosis. Patients with *Staphylococcus* osteomyelitis experience severe pain and limping. MRI accurately

localized the lesion, showed the extent, and helped plan the approach. All children were surgically treated with an appropriate open procedure, arthrotomy, and debridement, and were then treated with antibiotics according to the causative bacteria. They reported that a delay in surgical treatment could lead to joint destruction [5].

Houmami et al. reported primary epiphyseal subacute osteomyelitis caused by *Mycobacterium* species. They performed a systematic literature review and reported 16 children at their institution. They reported that *Mycobacterium* osteomyelitis has a mild course, with positive cultures in up to 50% of the surgical bone specimens. Delayed diagnosis despite surgical drainage may lead to complications, such as physeal damage, leg length discrepancy, or angular deformities [17].

St Jeor reported the case of a 6-year-old girl with multifocal subacute osteomyelitis, with small peripheral osteolytic lesions in the distal fibula and tibia in the ankle joint, without septic arthritis, which was aspirated with fluoroscopic guidance, and the cultures were negative. The patient was treated with oral antibiotics and resumed normal activities [18].

In contrast, Hamdy et al. and Ezra et al. favor the conservative treatment of subacute osteomyelitis with intravenous antibiotics. A report from Ezra described 55 children, wherein epiphyseal involvement was observed in 16 children. After conservative treatment, lesions were reported as cured 3-12 months, after the initial diagnosis. Despite an unclear initial diagnosis, they proposed conservative treatment [11, 12].

Dartnell et al. reported acute and subacute pediatric osteomyelitis in an instructional and systematic literature review. They favor the intravenous use of antibiotics in acute uncomplicated osteomyelitis that can be changed to oral antibiotics according to clinical and laboratory improvements. They recommend surgery if the child did not show improvement and for collection of pus, which increased the accuracy of bacteriological diagnosis. Surgery was performed for abscess collection as an urgent procedure. They reported that laboratory markers (CRP and ESR) remained elevated for a longer time in surgically treated children [14].

We believe that surgical treatment is essential for confirming the diagnosis of subacute osteomyelitis. An osteolytic lesion in the epiphysis, with possible extension in the metaphysis, requires diagnosis in a child with no clinical symptoms of acute osteomyelitis. With thorough cleaning of the joint, we provided chances for the best recovery, avoiding lesions of the articular tissue and the growth plate.

A positive blood culture is an exception as a result, from subacute osteomyelitis. Classic cultures were negative in three of our patients. The girl with purulent tissues had a PCR culture that was positive for *Staphylococcus aureus*. Green et al.reported in their series positive cultures only in two children [19].

In a study of 65 consecutive cases of primary hematogenous osteomyelitis over 16 years, two forms of bacteriological etiology have been reported. In the infantile form, between 6 months and 4 years of age, only *Kingella kingae* (*K. kingae*) were cultured, while in those older than 4 years, *Staphylococcus aureus* was the predominant bacteria. Using PCR assays, they reported 86% positive results and proposed the use of a PCR assay specific for *K. kingae* in children younger than 4 years [20].

We used antibiotics for *Staphylococcus aureus* in all our patients for 3 weeks. Based on the clinical improvement and normalization of laboratory markers, we stopped further use of antibiotics.

Subacute osteomyelitis may occur in children with sickle cells because of bone infarction. All patients were initially evaluated with appropriate blood tests, which were normal. In children, the most common causative bacteria are *Staphylococcus aureus* and *Salmonella*[14].

In conclusion, subacute epiphyseal osteomyelitis is a rare condition with obscure clinical and radiological findings. MRI provides the most accurate diagnosis. Surgical treatment with debridement and pathological examination was used to establish an accurate diagnosis. With surgical management, further lesions of the growth plate with subsequent limb deformities were avoided.

Author Contributions: NL, designed the study, wrote the original draft, reviewed and edited; CC and PK assisted in surgery, assisted in editing and reviewing the draft.

Ethics approval and consent to participate: The study has been approved from the editorial board and the ethic committee of our hospital. All parents of children have concerned for participating in the study.

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