Acute primary epiphyseal osteomyelitis of proximal tibia - A case report and review of the literature

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Learning Point of the Article:
Despite being a rarely reported entity, acute primary epiphyseal osteomyelitis warrants high suspicion and early diagnosis to prevent dreaded long term complications.

Abstract

Introduction: Acute pyogenic epiphyseal osteomyelitis is a rare condition, mostly affecting children. This case report discusses the importance of clinical suspicion and the role of adjuncts in the diagnosis of these cases.

Case Presentation: A 12-year-old female presented with pain in the right knee for 1 week and difficulty in walking for 4 days. Diagnosis was made with the help of magnetic resonance imaging MRI and guided biopsy. Pseudomonas aeruginosa was isolated from culture. She was treated with intravenous antibiotics with a good response.

Discussion: Despite being a rarely reported entity, primary epiphyseal osteomyelitis should be kept as one of the differentials of the painful swollen knee in a child. Early diagnosis with the help of proper radiological investigations is the key to prevent the sequelae of this disease.

Keywords: Epiphyseal osteomyelitis, Pseudomonas aeruginosa, tibia, children, epiphysitis, pyogenic osteomyelitis.

Introduction

Acute hematogenous osteomyelitis is common in children. It most commonly affects the metaphysis of long bones and typical clinical presentation makes diagnosis difficult to miss [1]. Primary osteomyelitis involving the epiphysis is rare, and the presentation is often atypical and vague, which may lead to missed or delayed diagnosis [2]. If treated appropriately, it usually follows a benign course in an otherwise healthy child. An acute or subacute form can set in regardless of age [3, 4]. Cases of epiphyseal osteomyelitis have previously been reported in the distal femur, proximal tibia, proximal femur, and proximal radius [5, 6, 7, 8, 9, 10]. We present a rare case of acute primary epiphyseal osteomyelitis of proximal tibia caused by Pseudomonas aeruginosa in a 12-year-old female with a review of the literature and various diagnostic modalities helpful in establishing an early diagnosis.

Case Report

A 12-year-old female presented to the outpatient department with pain in the right knee for 1 week and difficulty bearing weight for the past 4 days. The pain was insidious in onset without any preceding trauma and was aggravated with weight-bearing and knee bending. She had no constitutional symptoms. Her initial knee radiograph was normal (Fig. 1). Laboratory work-up showed a total leukocyte count of 12,700/mm³ with a
neutrophilic predominance, erythrocyte sedimentation rate of 29 mm3, and C-reactive protein of 5.28. Her serum Vitamin D was 12.3 ng/dl and other blood metabolic parameters such as liver and renal functional tests, serum calcium, phosphate, and alkaline phosphatase were normal. She was seronegative for HIV. Magnetic resonance imaging (MRI) showed hyperintensities in T2/STIR images in the epiphysis of the proximal tibia, more marked on the lateral aspect. There was a diffuse post-contrast enhancement, suggestive of edema (Fig. 1). Otherwise, the knee joint was unremarkable with no effusion or synovial hypertrophy. No local pus collection could be appreciated along the tibia. Ultrasonography-guided biopsy from the lateral tibial epiphysis was done and samples were sent for histopathological and microbiological examinations. Histopathological examination showed dense inflammatory infiltrate comprising neutrophils and lymphocytes, without any atypical or malignant cells. After 48 h of incubation, P. aeruginosa was isolated in the aerobic culture. Diagnosis of pyogenic proximal tibial epiphysitis was made. She was immobilized in an above knee POP slab and started on supportive measures including pain relief and nutritional care. No empirical antibiotic therapy was started. Culture-specific antibiotic treatment was started according to the antibiogram (intravenous ceftazidime 1000 mg thrice daily for 3 weeks followed by oral faropenem 200 mg twice daily for 3 weeks). Inflammatory markers were monitored at regular intervals. After 1 week of initiation of intravenous antibiotics, she improved clinically and inflammatory markers showed a decreasing trend. C-reactive protein was 2.0 and 1.7 at day 7 and 14 of treatment with antibiotics, respectively. She was completely pain free on day 12 and was started on passive knee range of motion, isometric quadriceps strengthening exercises, and partial weight-bearing with support. At 24 months follow-up, the child was active with knee range of motion 0–145 degrees of flexion and normal plain radiograph (Fig. 1). No local growth disturbance was noted.

The estimated incidence of acute primary pyogenic osteomyelitis is 0.5–1 in 5000 cases [1]. However, the incidence is constantly increasing due to more virulent organisms and better diagnostic modalities. Primary pyogenic osteomyelitis usually involves the metaphysis of long bones [2, 3] and various hypotheses have been proposed for the same in the literature. However, the isolated involvement of epiphysis is rare. Only a few cases have been reported in the literature so far (Table 1). Primary epiphyseal osteomyelitis was thought to be most commonly affecting children <3 years [12] but it can involve any age. The predisposing factors are poor host immunity, malnutrition, and overcrowding. The predominant site being the distal femur or proximal tibia (Table 1). The most common isolated organism being Staphylococcus aureus [2]. P. aeruginosa has been reported in few cases of metaphyseal osteomyelitis in adults mostly as nosocomial or chronic infection [13], but this reports a unique case of primary acute pyogenic epiphyseal osteomyelitis due to community-acquired P. aeruginosa infection in an otherwise healthy child. The usual presentation is with only mild-to-moderate local signs and symptoms and minimal or absent systemic manifestation which makes the diagnosis difficult [14]. Febrile episodes are seen only in 60–66% of children [15]. Long-term prognosis is highly favorable without any growth arrest if timely managed, although a delayed diagnosis can result in septic arthritis and destruction of growth plate [16]. MRI is more sensitive in diagnosing and localizing the site and extent of the lesion in primary epiphyseal osteomyelitis (sensitivity: 85–100% and specificity: 75–100%) with the earliest changes detected as early as 2–5 days following the onset of symptoms [17]. However, infection and inflammation cannot be differentiated. Nuclear imaging techniques such as three-phase bone scanning (sensitivity – 85–92% and specificity – 54–87%), gallium-67 citrate scanning (sensitivity – 81–100% and specificity – 40%), and indium 111 labeled leukocytes scintigraphy (sensitivity – 100%, specificity – 70–95%, and accuracy – 86%) are also helpful. False-negative bone scans are obtained if performed in early course of infection.
due to increase in medullary pressure in the lesion and in neonates [18, 19, 20]. Nuclear imaging techniques are also helpful in multifocal disease. Higher sensitivity and specificity of MRI in the early diagnosis of acute epiphyseal osteomyelitis makes it the imaging modality of choice. Ultrasonography is sensitive in detecting joint effusion and can obtain guided specimen from the epiphysis, without exposure to radiation. Bone biopsies and aspirates are culture positive in 31–83% of the cases, so negative growth does not rule out infection [21]. The mainstay in the treatment of primary pyogenic epiphyseal osteomyelitis is timely diagnosis and initiation of antibiotics according to the antibiogram [22]. Most cases respond to the

### Table 1: A literature review of articles describing acute pyogenic epiphyseal osteomyelitis

| S. No. | Author | Year | No. of cases | Age | Site | Symptom duration | Plain radiograph | Culture | Management | Follow-up | Outcome |
|--------|--------|------|--------------|-----|------|------------------|------------------|---------|------------|-----------|----------|
| 1      | Hwang et al. | 2016 | 1 | 12 years | Distal tibia | 9 days | Lytic lesion | Joint fluid and tissue – S. aureus | 1. Arthroscopic debridement followed by open debridement curettage and irrigation. 2. IV antibiotics for 3 weeks followed by oral antibiotics | 18 months | Normal |
| 2      | Green et al. | 1981 | 8 (5) | 2–4 years | Proximal tibia-2, Distal femur-2, Proximal femur-1. | N/A | N/A | N/A | 1. Curettage and irrigation 2. Antibiotics | N/A | N/A |
| 3      | Rosenbaum and Blumhagen | 1985 | 9 (7) | 21 months–9 years | Distal femur-6, Proximal tibia-1, Proximal humerus-1. | 1–28 days | Lytic lesion | Blood culture- 1 case positive for S. pneumoniae 2)Tissue culture–2 positives (1 – S. aureus, 2 – H. influenza) | 1. Arthrocentesis-5 cases 2. IV antibiotics followed by oral antibiotics | N/A | N/A |
| 4      | Abdelgawad et al. | 2007 | 1 | 17 months | Distal femur | 6 days | Normal | 1. Blood – negative. 2. Tissue culture– Salmonella Typhi B | 1. Fluoroscopy-guided abscess drainage – thrice done 2. IV antibiotics for 4 weeks | N/A | Normal |
| 5      | Sorensen et al. | 1988 | 3 | 20 months | Proximal tibia | 14 days | Lytic lesion | Tissue culture – negative | 1. Curettage and bone grafting 2. IV antibiotics for 10 days | 9 months | Normal |
| 6      | Rasool | 2001 | 2 | 2–12 year, Mean-7.5 years | Proximal and distal femur | Mean-2 weeks–3 months | Lytic lesion | N/A | 1. Fluoroscopic guided extra-articular biopsy and curettage 2. IV antibiotics for 4–5 days followed by oral antibiotics for 6 weeks 3. Immobilization for 4–6 weeks | Mean 2.4 years | Normal |
| 7      | Maffulli and Fuxsen | 1990 | 1 | 6 years | Proximal radius. (Epiphysitis with septic arthritis) | 2 months | Lytic lesion | Tissue culture – negative | 1. Curettage and irrigation 2. IV antibiotics for 1 week and oral antibiotics for 6 weeks | 10 months | Normal (Reossification of radial head) |
| 8      | Longjohn et al. | 1995 | 2 | 4 years 2 months | Lateral femoral epiphysis | 2 weeks | Lytic lesion | Blood culture – S. aureus. Tissue culture – S. aureus | 1. Curettage and irrigation 2. IV antibiotics for 4 weeks and oral antibiotics for 10 days | 6 years | Normal |
| 9      | Kramer et al. | 1986 | 1 | 11 years | N/A | 6 days | N/A | Blood culture – Neg Joint fluid – S. aureus | N/A | N/A | N/A |
| 10     | Kao et al. | 2003 | 2 | 28 months | Distal femur | 10 days | Lytic lesion | Blood and joint aspirate – Neg Tissue – Salmonella enteritidis | 1. Curettage 2. IV antibiotics for 5 weeks | 16 months | Normal |

Footnote: The numbers mentioned in brackets are the actual number of cases of isolated epiphysitis, Neg: Negative, N/A: Not available, S. aureus: Staphylococcus aureus
antibiotics but if there is no response to antibiotic treatment, surgical intervention is indicated in the form of curettage, aspiration, and debridement.

**Conclusion**

Despite being a rarely reported entity, primary epiphyseal osteomyelitis should be kept as one of the differentials of the painful swollen knee in a child. Early diagnosis with the help of proper radiological investigations is the key to prevent sequelae of this disease.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient’s parents have given their consent for patient images and other clinical information to be reported in the journal. The patient’s parents understand that his names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil Source of support: None

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**Clinical Message**

Although historically primary epiphyseal osteomyelitis ran a benign course, soon it may follow a more aggressive course due to increased virulence and antibiotic resistance in organisms. Therefore, high suspicion of infection is warranted and diagnostic modalities should be appropriately used without sticking to a single modality. Early diagnosis and initiation of treatment prevent long-term sequelae and crippling.
Conflict of Interest: Nil
Source of Support: Nil

Consent: The authors confirm that informed consent was obtained from the patient for publication of this case report

How to Cite this Article
Das L, Goyal T, Paul S, Gupta T. Acute Primary Epiphyseal Osteomyelitis of Proximal Tibia – A Case Report and Review of the Literature. Journal of Orthopaedic Case Reports 2021 December;11(12): 52-56.