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Case Reports

Epiphyseal Cystic Tuberculosis: A Case Report and Review of Subject

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CASE REPORT

A boy of six years, presented in the out patient department with pain right knee for the last four months and limp while walking. The pain in the knee started gradually, it was mild to begin with and gradually progressed over the passage of time. The parents noted limp on walking. The limp became more obvious during last four weeks. There was no history of trauma to knee, no history of fever or swelling of knee. There was no history of cough, expectoration, weight loss. There was nothing significant in the past history. He did not take any medications. He belonged to a poor social class.

On examination, he was of thin built. He had minimal limp on walking, with an antalgic gait. The examination of chest, cardiovascular system, central nervous system, and abdomen was unremarkable. Local examination of knee revealed mild swelling of knee. There were no scar marks. The medial condyle of right femur was more prominent as compared to left side. There was no tenderness on deep palpation. There was no quadriceps wasting and range of movements, both active and passive at right knee was normal. Hip examination showed mild degree of reduction of internal rotation.

The radiological examination of right knee revealed mild soft tissue swelling, there was an irregular radiolucent area in the medial aspect of distal epiphysis of right femur with patchy calcification, limited to medial condyle (Fig. 1). The x-ray of chest was normal. The routine laboratory test were carried out. The hemoglobin was 11.1 gm/dl, WBC 7900, and ESR 22 mm first hour.

A provisional diagnosis of chondroblastoma of right distal femur was made and biopsy planned. Under general anesthesia and pneumatic tourniquet, anteromedial approach was used and cortical window made through epiphysis. Lytic lesion was found, filled with purulent fluid and sequestra, which was aspirated and sent for culture sensitivity, Gram staining, and AFB. Curettage of lesion was performed, multiple small sequestra came out. The cavity was irrigated and sequestra sent for histopathology. There was no involvement of synovium or articular cartilage. The wound repaired over suction drain and plaster cast applied. The post operative recovery was smooth and uneventful.

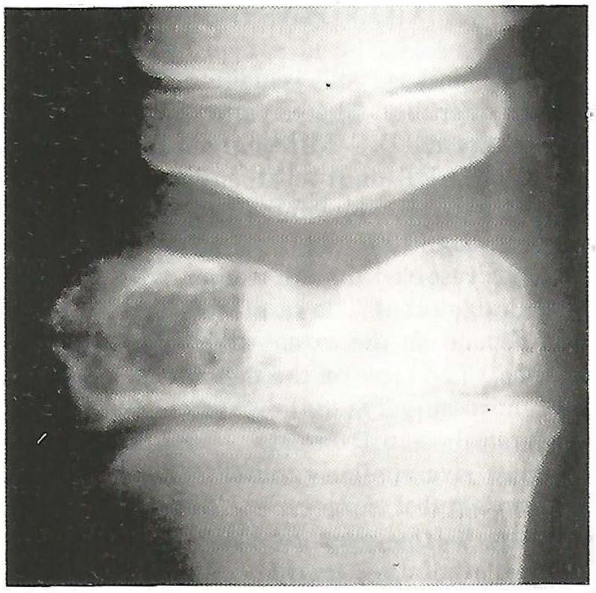

Fig. 1: X-rays of knee joint showing radio lucent area in distal femoral epiphysis.

The microbiological examination with Gram staining revealed multiple pus cells however, there were no Gram positive or Gram negative micro-organisms. The Ziehl Neelson stain did not reveal any acid fast bacilli. The routine culture of smear did not show any growth after 48 hours.

The histological examination revealed multiple grayish white partly cartilaginous and partly bony pieces of tissue measuring 2.5 cm in aggregate. On microscopy, the section showed fibro-cartilaginous tissue, cartilage, bony trabeculae containing focal areas of necrosis, dead bone, focal suppuration and heavy chronic inflammatory cell infiltrate of many plasma cell, lymphocytes, polymorphs and granulation tissue. In addition, there were many epithelioid granulomas, some with central necrosis

and few Langhan's type of multinucleated giant cell. The appearances were of chronic granulomatous inflammatory lesion of bone, most likely tuberculosis. There was no evidence of malignancy.

The patient was started on anti-tuberculous therapy and he is being followed up in the out patient department.

REVIEW OF SUBJECT

Tuberculosis remains a major cause of skeletal infection in under-developed and developing countries. It is a chronic infection caused by Mycobacterium tuberculosis. It has been reported that musculoskeletal involvement occurs in about one percent of cases of tuberculosis, although the incidence is higher in some populations[1]. The disease is usually secondary to a primary focus which is usually in the thoracic cavity or gastrointestinal tract; in 75 percent patients, primary focus is in the lungs[2]. Goldblatt and Cremin[3] found chest involvement on x-rays only in 31 percent of cases. The organism reach the synovial joint by haematogenous spread or by direct extension from an infective focus in the adjacent bone. If spread occurs from an adjacent bone, there is continued enlargement of tuberculous abscess with bony destruction, which eventually ruptures into joint. When this occurs, the destruction is more rapid than in haematogenous form[4].

The synovial involvement in tuberculosis of joints is early. It leads to effusion and synovial thickening. There is gradual destruction of articular cartilage by the tuberculous granulation tissue. The caseous necrotic tissue forms cold abscess which spreads through facial planes and erupts externally. The additional bacterial infection exaggerates the destructive process[5]. Fifty percent of cases show vertebral involvement, 15 percent hip, 20 percent ankle, elbow and other bones[6,7].

Tuberculus involvement of bones in the form of multiple cystic tuberculosis is a well known entity but solitary cystic lesion are uncommon and not well documented[8]. Lesions in the bones in disseminated form are usually seen with in three to six months and start at the time of initial bacilemia, where as cystic bone lesions occur about one year after initial infection[9].

Cystic tuberculus involvement of epiphysis is very rare. We searched the literature using CD-ROM Medline from 1966 to 1994. We were able to find only a few references on this topic. We feel that less than a hundred cases of epiphyseal tuberculosis would have been reported including those caused by BCG vaccination.

Epiphyseal involvement in tuberculosis may be haematogenous, secondary to joint involvement where the lesion usually starts from the periphery.The epiphyseal tuberculosis may develop after BCG vaccination. The knowledge of the complication, of osteomyelitis after vaccination, is small although the first case was published more than 40 years ago. The disease is seen in the first five years of life in otherwise healthy children. Most cases are localized near the epiphysis of the long bones[10]. A localization in the spine has also been published. The highest frequency of BCG osteomyelitis is reported in Finland and Sweden with about 40 cases per 1 million vaccinations.

In 1973 an increase in the number of osteitis caused by BCG vaccination in the neonatal period was observed in Sweden[11]. A high incidence of this complication, i.e. 1 case per 3000 vaccinated newborn children continued until this vaccination was interrupted in 1975. Of the total of 152 known cases in Sweden, 82 were boys and 70 girls. The mean incubation period was 14 months for boys and 23 for girls. Extremely long incubation periods were seen, i.e. 6 years in two children and 12 years in one. Eleven children had multiple osteitis lesions. The epiphyses of the long bones of the extremities were the most frequent sites of the affection (109 lesions). Only in 6 patients the spine, which is the common site of osteitis caused by tuberculosis, was affected. Relapses occurred in two patients.

Peltola-H et al[12] reported ten patients with tuberculous osteomyelitis and three with a subcutaneous abscess, all caused by BCG vaccination. All patients were less than 3 years old. The sites of predilection of osteomyelitis were the metaphysis or epiphysis of the femur.

Cystic tuberculosis of the bones is a recognized entity effecting long and flat bones either multiple or solitary lesions but only a few case have been reported effecting epiphysis alone. The disease is usually haematogenous[2,13-16].

Jungling[17] in 1920 described multiple cystic lesions of bone confined to hand and feet and termed them osteitis tuberculosa multiplex cystoides or Junglings Disease. Tuberculin test was negative. No acid fast bacilli were isolated and no caseation

necrosis was found. Later Ellis[18] classified these lesions to be from Boecks Sarcoidosis. Brian O'connor[19] reported on criteria for confirmation of diagnosis as presence of caseation necrosis plus granulomas of tuberculosis and presence of acid fast bacilli.

In adults, common sites are axial skeleton shoulder and pelvic girdle[9,20]. In children, metaphysis of long bones are common site possible due to more vascularity in this region[2]. Bone lesions remain solitary usually because of sensitization to tubercle bacilli. If host immunity is low then multiple lesions are produced[8].

The radiolucent epiphyseal lesion is a dilemma for diagnosis[21]. In region of hip, it can be easily confused with Perthes disease[22,23] and septic arthritis[24]. Gardner-DJ and Azouz-EM[25] evaluated retrospectively the varying radiographic appearances of 15 solitary lucent epiphyseal lesions occurring in children. Imaging modalities used included plain films, conventional tomography, nuclear scintigraphy, and computed tomography. Forty percent of the lesions (6) were due to osteomyelitis. The remaining lesions included tuberculosis (1), foreign body granuloma (1), chondroblastoma (2), chondromyxoid fibroma (1), enchondroma (1), osteoid osteoma (2), and eosinophilic granuloma (1). Although the radiographic appearances of such lesions may be particularly characteristic, pathologic correlation is frequently necessary. The high incidence of osteomyelitis in this study emphasizes its importance as a cause for a lucent epiphyseal lesion.

Lebarbier-P et al[26] reported four cases which all showed at first almost the same radiographical features : an epiphyseal defect of the knee. The first case developed in an inflammatory context and proved to be an osteomyelitis. The second case showed a benign tumor on X-Ray examination a chondroblastoma. Then an epiphyseal tuberculosis was produced which at first led to a rather unsettled diagnosis. The last case was a rare observation of chondroma. They felt that diagnostic problems are frequent and besides possible infections localizations, the diagnosis of chondrablastome seems to be the most likely. They recommended surgical approach of such epiphyseal lesions, not only to corroborate etiology but also in order to avoid an increase in the volume that could impair the epiphyseal plate endanger the growth.

Versfeld et al[27] reported that radiological features of joint tuberculosis are non specific and include soft tissue swelling slow destruction of cartilage, peripheral erosive defects, periarticular atrophy (demineralization). Minimal periosteal reaction, slow enlargement of focus and involvement of adjacent joint, abscess formation. Late feature is soft tissue calcification. Epiphyseal plate and joint offer little resistance to spread. Diagnosis is confirmed by histological analysis.

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