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Research Article

ISOLATED CYSTIC TUBERCULOSIS OF BONE IN CHILDREN: A DIAGNOSTIC DILEMMA

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ABSTRACT
Cystic tuberculosis is a major cause of morbidity and mortality in children. The radiographic appearance of osteoarticular tuberculosis can mimic metastatic tumors or some primary osseous lesions, such as eosinophilic granuloma, especially if multiple destructive lesions are present. The objective of this study was to highlight the diagnostic difficulties that bone and joint tuberculosis poses for clinicians and other persons involved in health care delivery system.

Key Words: *Cystic bone tuberculosis, Anti-tubercular therapy, Curettage.*

INTRODUCTION
Osseous tuberculosis is a major cause of morbidity and mortality in children. Cystic tuberculosis of the bone is a rare form of tubercular osteomyelitis associated with disseminated lesions. Resnick (1995) stated that the multiple cystic forms was first described in 1920 as 'Jung ling's disease' or 'osteitis tuberculosa multiplex cystoides. The bone lesions in tuberculosis are radiolucent, round to oval, and are situated in the peripheral skeleton near the metaphysis. The cyst-like appearance is due to the variable marginal sclerosis. These 'cysts', however, can occur anywhere in the skeleton. Solitary lesions may mimic bacterial and fungal infections, simple or aneurysmal bone cysts, cartilaginous tumors and osteoid osteoma. Muradali *et al.*, (1993) described that skeletal tuberculosis is thought to occur secondary to lymphohematogenous dissemination to the skeleton at the time of initial pulmonary infection. Reider (1990) found that there may be no radiographic evidence of pulmonary involvement in about 50% of patients. Alvarez (1984) described that Clinical and radiographic presentation of skeletal tuberculosis in patients from endemic areas differs from that of individuals from nonendemic areas. Patients from endemic areas present with a higher incidence of multifocal skeletal involvement. Radiographic features may present as periosteal reaction, bone sclerosis, and severe bone destruction. The profile from nonendemic areas is of an older patient, usually with a debilitating underlying disease; lesions are usually solitary, osteolytic, and involving the axial skeleton, thoracolumbar vertebral bodies, and hips. Here by we are reporting 16 cases of cystic tuberculous bone disease, diagnostic dilemma and management.

MATERIALS AND METHODS
This prospective study was carried out at department of Orthopaedics in M.M.Medical College from June 2005 to June 2010. Institutional medical ethics committee approved it. 16 patients (<12 years of age) with cystic bone tuberculosis admitted to our institute were included. All the children were treated by surgical debridement and curettage followed by anti-tubercular chemotherapy. All children were followed for twenty four months.

Only biopsy-proven cases were included in this study. A written informed consent was obtained from all the patients; they were explained about treatment plan, cost of operation, and hospital stay after surgery, and complications of anaesthesia. To rule out any co-morbid conditions, haematological tests including haemogram, erythrocyte sedimentation rate, peripheral blood smear, blood sugar levels, liver and kidney function, hepatitis profile, and human immunodeficiency virus, according to the protocol for diagnosis and management. The Mantoux skin test was done in all patients. Radiographs of the chest (including shoulders), whole spine, pelvis (with both hips), knees, elbows, hands, and feet were taken. Ultrasonography of the abdomen was performed for detection of visceral involvement. The diagnosis was

confirmed histologically through sputum sample, pus (for Zeil-Neelson staining), ulcer edge biopsy, curetting from the soft-tissue collections, and biopsy from bony lesions and regional lymph nodes depending on site of involvement. 4-drug antitubercular chemotherapy (isoniazid, rifampicin, ethambutol, pyrazinamide) for 2 months, followed by a 2-drug regimen (isoniazid and rifampicin) for 10 months was given to histopathology positive patient. They were followed up after surgery, were clinically and radio logically assessed for defect healing and joint movements. Treatment progress was monitored using serial haematological tests and radiographs at 2, 5, 8, and 12 months. According to the criteria the results was graded as--

Excellent **-** when the lesions heal within 16 weeks without any complication

Good **-** healing occurs within 24 weeks with treatable complications like joint stiffness

Poor - when healing occurred before or after 24 weeks with one or more permanent complications like limb shortening and permanent joint stiffness.

Children were followed for 24 months.

RESULTS AND DISCUSSION

All the 16 children (12 boys and 4 girls) with a mean age of 5 years (1 – 12 years) were treated by surgical debridement and curettage followed by anti-tubercular chemotherapy. All children were followed for twenty-four months. The duration of symptoms ranged from 1 week to 3 months. Seven children had a history of mild trauma. The symptoms and signs, which included pain and swelling, were related mainly to the joint adjacent to the bone involved. Ten children had antalgic gait or limping due to involvement of lower limbs. Nine were diagnosed having pyogenic osteomyelitis that underwent surgical drainage shortly after admission. The diagnosis of tuberculosis was made subsequently. Two patients presented with sinus formation, out of this second patient had a discharging left iliac bone sinus and patient 5 had multiple sinuses in right humerus. In addition, another two cases had dactylitis of the thumb and ring finger. Four patients had multiple cystic lesions and twelve had solitary cystic lesions. Two patients had metatarsal bone lesion and one had in calcaneum. Loss of weight and appetite, fever and cough were not the predominant features. Chest involvement was seen in two children. The ESR, full blood count and the Mantoux skin Chest radiographs and bone scans were done. The ESR ranged from 46 mm/hr to 95 mm/hr in eleven children; in five, it was normal. The hemoglobin level ranged from 8.5 gpercentage to 12.6 gpercentage and the white cell count from 5.6 x 10ı/l to 13.8 x 10ı/l. The Mantoux test (M. T.) was positive in nine children and negative in seven. In radiological findings, four of the children had multiple cystic lesions and thirteen had solitary cystic lesions. (Table I). The total number of lesions in all the patients was 21, including nine in the metaphyses of long bones, three in the epiphyses and three in the diaphysis. The other lesions were in the hand (2) pelvis (1), foot (2), and calcaneum (1). The cysts were between 0.5 cm and 3 cm in diameter. Localized osteoporosis was seen in all the patients. Sequestrum formation was seen in two patients (5, 12); the dense sclerosis around this sequestrum resembled the appearance of an osteoid osteoma. In metaphyseal lesions, these were typically expanding, radiolucent and round to oval with a variable amount of sclerosis. There was a multilocular appearance in four cases. Mild periosteal reaction occurred in five cases, dense periosteal reaction around the subtrochanter and fibula was observed in two (12, 15). Lesions in the distal tibia, proximal tibia and proximal humerus were similar to aneurismal and simple bone cysts. In three patients the growth plate was transgressed and the lesions involved the epiphyses. In case 3, (Figure 1, 2 and 3) the appearance resembled a chondroblastoma. Epiphyseal lesions - Primary epiphyseal involvement was seen in only three patients as small punched-out areas with marginal sclerosis. In diaphyseal lesion - the right shaft of humerus, right shaft of fibula and left subtrochanteric regions were involved in case 5 (multicystic),12 (multicystic) and 15 (Unilocular). In hand- In patient no.13, Left thumb at proximal phalanx at base showed unilocular lesion. Patient no 14 - ring finger at proximal and middle phalanx showed unilocular round and honeycombed, round lesion. In foot-- in patient 6, right calcaneous showed unilocular oval lesion. In case 9, the Base of 1 and 2 metatarsal of left side showed honeycombed, round and unilocular oval lesions. In

case 16, the Right great toe at metatarsal head showed unilocular oval lesion. In Pelvis - in case 2, the left iliac bone near iliac crest showed single punched out lesion. In joint involvement, in cases 1, 3, 7,8,9,13,14 and 16, the adjacent joint was involved in these patients with epiphysis or Metaphysical lesions. All showed soft-tissue swelling, osteoporosis and joint-space widening. In all patients, the diagnosis of tuberculosis was confirmed histologically. Care was taken to obtain biopsy specimens from the area of bone destruction rather than from the synovium of the adjacent joint. In each case, the cyst-like cavity was curetted to remove all granulation tissue and pus; bone grafting was done in case 3 to fill the resultant defect. After surgery, the affected limb was splinted in plaster for four weeks, using a bilateral above-knee abduction plaster for the patient with hip involvement. Active physiotherapy was started after the removal of the splint. Patients were treated with standard 4-drug antitubercular chemotherapy (isoniazid, rifampicin, ethambutol, pyrazinamide) for 2 months, followed by a 2-drug regimen (isoniazid and rifampicin) for 10 months. Follow-up was from 6 months to 2 years in 16 children. All patients (except cases 1, 5, and 7) had a full range of painless movement of the adjacent joints at the end of medical treatment. Case 1 had a fixed flexion deformity of 20 degree at knee joint, Case 5 had a fixed flexion deformity of 30 degree at right elbow joint. Patient no 7 had a fixed flexion deformity of 30 degree at right hip joint. In these three patients, the radiological defects had healed well. In the remaining 13 patients, the radiological defects healed completely in five cases and in eight cases defects healed with sclerosis. There was evidence of growth disturbance in two patients, with underdevelopment of the first metatarsal in case 9 and of the right femur in case 7. Remodeling of expanded lesions occurred in all the patients. The results were excellent in 87.5% and good in 12.5% patients. Out of sixteen children, the radiological defects healed with sclerosis in eight children. In rest 50%, the radiological defects healed completely. There was evidence of growth disturbance in two patients, with underdevelopment of the first metatarsal in case 9 and of the right femur in case 7. Remodelling of expanded lesions occurred in all the patients. All children had a full range of painless movement of the adjacent joints at the end of medical treatment. The results were excellent in 87.5% and good in 12.5% patients.Complications was found in five patients. Tuberculosis remains the major source of morbidity and mortality worldwide, affecting approximately one-third of the world's population. Osteoarticular involvement occurs in less than 3% of patients with extra pulmonary tuberculosis and of which spine represents half of these lesions. Ryan (1930) described that the tuberculosis of the ilium is a rare identity, and until now fewer cases are reported in literature. In my study, one case of solitary cystic lesion of left ilium was seen in 12 years old girl having draining sinuses for the last few months. Tuberculous infection of metacarpals, metatarsals and phalanges of hands and feet is known as tuberculous dactylitis. Kushwaha (2008) stated that Bone and joint tuberculosis occur in 1-5% children who have untreated initial pulmonary tuberculosis and spread to the skeletal system during the initial infection via the lymphohaematogenous route. In my study, two children had pulmonary tuberculosis. Gyanshankar (2009) stated that the skeletal infection becomes symptomatic within 1-3 years after the initial infection. 85% of children with tuberculous dactylitis are younger than 6years of age and its incidence among children with tuberculosis was reported to be 0.65%-6.9 %. The condition usually presents as a painless swelling of a digit of a few months duration. The radiographic feature of cystic expansion of short tubular bones has led to the name of spina ventosa being given to tuberculosis dactylitis of the short bones of the hand. Periosteal reaction and sequestra are not common but may occur. Sclerosis may be seen in long standing cases. In my study three cases (18.75%) of tuberculous dactylitis were found, in which two cases (12.5%) in hands and one case (6.25%) in foot were seen. There was slight shortening of first metatarsal of left foot but healing occurred completely with sclerosis. Four children (25%) had multiple cystic lesions and twelve (75%) had isolated lesions. Most patients present with fever, soft tissue swelling and pain at the involved site. Though sinus formation is reported to be rare by Rasool (1994) and Versveld (1982), Seth (2001) reports multiple sinuses or sinus tracts to be a common presentation. We too had two cases who presented with sinus formation. Low-grade fever, anorexia and weight loss are commonly seen and muscle atrophy around the joint is an early predominant feature. Tubercular osteitis is usually the result of hematogenous

dissemination, investigations should be aimed at finding the primary site of infection. Shannon (1990) has been stated that in tuberculosis of the bone, the primary lesion is usually in the chest and that the tuberculin reaction is always positive. Among our patients, only nine had a positive Mantoux test and only two had evidence of pulmonary involvement. Cytopathology obtained by FNAC and/or biopsy of the bone or synovium is recommended for diagnosis and helps rule out other conditions. Kher (1993) and Kothari (1999) report that the differential diagnosis of multiple osteolytic lesions mimic a variety of childhood malignant conditions like histiocytosis, lymphoreticular malignancy, syphilitic, pyogenic or mycotic osteomyelitis or neuroblastoma. Single lesions may be confused with sickle cell anemia, syphilitic lesions, leukemia, hyperparathyroidism or fibrous dysplasia. Anti-tubercular therapy remains the mainstay of treatment and results in a dramatic response in 85% cases when given for a period of nine months to a year. In 15% cases, with avascular lesions or lesions with caseation or sequestration, chemotherapy alone is not enough. In such situations, excision of the diseased focus increases the vascularity and allows the antitubercular drugs to reach the site of the lesion. Curettage of the cyst-like cavity to remove all the granulation tissue and pus, with or without bone grafting has been recommended. Persistently draining sinuses, which are infected, require broad-spectrum antibiotics in addition. Prognosis is good as response to antituberculous therapy is dramatic and only 15 % cases require surgery. Significant radiological improvement is seen as early as six weeks. Rafiqi (2013) stated that rare osteoarticular tuberculosis locations often cause diagnostic problems. Any chronic clinical presentation or suspected atypical bone lesion should suggest a diagnosis of osteoarticular tuberculosis. Rasool (2013) stated that the variable radiological picture of tuberculosis of the hip region can mimic various osteoarticular conditions in children. Biopsy is essential and should be taken from the bony lesion and not the synovium alone.

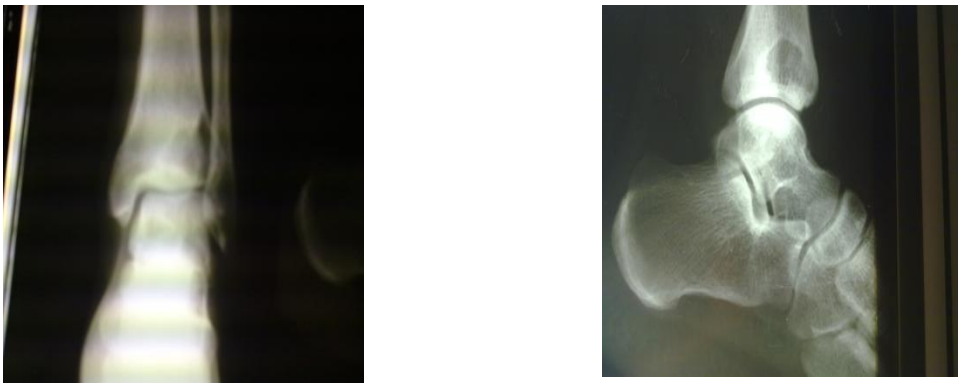

Figure 1: Radiograph of AP & lateral view of right ankle showing cystic lesion over lateral aspect of lower end of tibia.

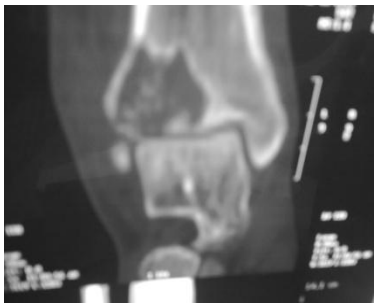

Figure 2: CT scan of lower end of right tibia showing cystic lesion communicating with the ankle joint.

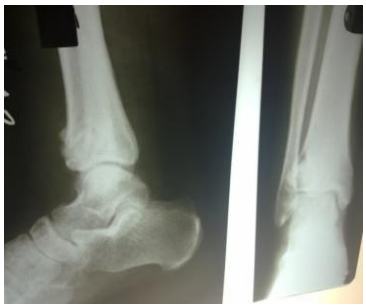

Figure 3: Post operative radiograph of right ankle showing cavity filled with cancellous bone graft.

Table1: Details of 16 children with cystic tuberculosis of bone

Case	Age (years)/Sex	Chief complaints	Anatomical site	Radiological findings	Investigation	Diagnosis	Follow-up (YEARS)	Healing	Joint movement
1	12/m	Soft tissue swelling, pain	Distal femoral epiphysis, proximal tibial metaphysis around knee joint.	Multiple Cystic lesions in femur and tibia.	M.T negative, fine needle aspiration cytology (FNAC) bone positive	Multiple cystic TB of bone	2	complete	Fixed flexion deformit Y 20 degree
2	12/F	Soft tissue swelling, pain, draining sinuse	Left iliac bone	Single punched out lesion in the iliac bone.	M.T negative, FNAC bone Positive.	solitary cystic lesion in iliac bone	2	complete	Normal function of iliopelvic joint.
3	12/M	Soft tissue swelling, pain	Lower end of tibia at metaphysis and epiphyseal region.	Unilocular oval	M.T negative, FNAC bone positive	solitary cystic lesion IN LOWER END OF TIBIA.	2	Sclerosis.	Normal range of motion.
4	10/F	Soft tissue swelling, pain	Upper end of tibia at metaphysis	Unilocular oval	M.T. POSITIVE. FNAC bone Positive	Solitary cystic lesion in upper end of tibia.	2	Sclerosis	Normal range of motion
5	7/M	Swelling, pain, multiple draining sinuses.	Right humerus At metaphysic and diaphysis.	Multiple cystic Lesions.	M.T. POSITIVE. FNAC bone Positive	Multiple cystic Lesions in right humerus.	2	Sclerosis	Fixed flexion deformit Y 30 degree

6	5/M	Soft tissue swelling, pain	Right calcaneum	Unilocular oval	M.T. POSITIVE. FNAC bone Positive	Unilocular oval in right calcaneum	2	Sclerosis	Normal range of motion
7	6/F	Soft tissue swelling, pain	Head (epiphysis) neck (metaphysic) of femur and acetabulum of right side.	Multiple cystic Lesions.	M.T. POSITIVE. FNAC bone Positive	Multiple cystic Lesions in Head neck of femur and acetabulum of right side.	2	Complete Slightly short femur	Fixed flexion deformit Y 30 degree
8	5/M	Soft tissue swelling, pain	Radial head and neck left side	Unilocular oval	M.T. POSITIVE. FNAC bone Positive	Unilocular oval IN Radial head and neck left side	2	complete	Normal range of motion
9	2/m	Soft tissue swelling, pain	Base of 1 and 2 metatarsal of left side.	Honeycombed, round and Unilocular oval	M.T. POSITIVE. FNAC bone Positive	Honeycombed, round and Unilocular oval in base of 1 and 2 metatarsal of left side.	2	Complete First metatarsal found slightly short.	Normal range of motion
10	3/F	Soft tissue swelling, pain	Lower end of femur at metaphysis	Unilocular oval	M.T negative, FNAC bone positive	Unilocular oval in Lower end of femur at metatarsal	2	Sclerosis	Normal range of motion
11	4/M	Soft tissue swelling, pain	Left head of humerus at metaphysic.	Unilocular oval	M.T. POSITIVE. FNAC	Unilocular oval in left head of	2	complete	Normal range of motion

					bone Positive	humerus at metaphysic			
12	10/M	Soft tissue swelling, pain	Lower end of metaphysis and diaphysis of fibula bone right side.	Multiple cystic Lesions	M.T negative, FNAC bone positive	Multiple cysticLesions in lower end of metaphysis and diaphysis of fibula bone right side.	2	Sclerosis	Normal range of motion
13	7/M	Soft tissue swelling, pain	Left thumb at proximal phalynx at base.	Unilocular round	M.T. POSITIVE. FNAC bone Positive	Unilocular round in Left thumb at proximal phalynx at base	2	complete	Normal range of motion
14	8/M	Soft tissue swelling, pain	Right ring finger at proximal and middle phalynx.	Unilocular round and Honeycombed, round	M.T negative, FNAC bone positive	Unilocular andHoneycombed, round in right ring finger at proximal and middle phalynx.	2	Sclerosis	Normal range of motion
15	5/M	Soft tissue swelling, pain	Left subtrochanteric region at diaphysis	Unilocular oval	M.T.negative,FNAC bone positive	Unilocular oval in left subtrochanteric region	2	Sclerosis	Normal range of motion
16	12/M	Soft tissue swelling, pain	Right great toe at metatarsal head	Unilocular oval	M.T.POSITIVE.FNACbonepositive Positive	Unilocular oval in right great toe at metatarsal head	2	complete	Normal range of motion

Conclusion

We conclude that the diagnosis of pediatric skeletal tuberculosis can be made with good correlation of clinical, radiographic, and laboratory findings. Biopsy and culture are the gold standards in diagnosis. Prognosis is good with chemotherapy and non-operative management. Surgical intervention may be needed in selected cases.

REFERENCES

Resnick D, Niwayama G (1995). Osteomyelitis, septic arthritis and soft tissue infections: Organisms. In: *Diagnosis of Bone and Joint disorders* **4** (3) 2448-2558.

Muradali D, Gold WL, Vellend H, et al(1993). Multifocal osteoarticular tuberculosis: report of four cases and review of management. *Journal* of *Clinical Infectious Diseases* **17**(2)204 -209.

Reider HL, Snider DE Jr, Cauthen GM (1990). Extra pulmonary tuberculosis in the United States. *American Review of Respiratory Disease* **141**(2)347-351.

Alvarez S, McCabe WR (1984). Extrapulmonary tuberculosis revisited: a review of experience at Boston City and other hospitals. *Medicine* **63**(1)25-55.

Ryan CA, Funston RV (1930). Osteomyelitis of ilium (probably tuberculous): case report. *Journal Bone & Joint Surgery* [Am] **12**(1)165-167.

Kushwaha RA, Kant S, Verma SK, Sanjay, Mehra S (2008). Case report: Isolated metacarpal bone tuberculosis-A case report. *Lung India* **25** (1)17-9.

Gyanshankar PM, Dhamgaye TM, Amol BF(2009). Spina ventosa discharging tubercle bacilli--a case report. *The Indian Journal Tuberculosis* **56** (2)100-3.

Rasool MN, Govender S, Naidoo KS (1994). Cystic tuberculosis of bone in children. *Journal of the Bone and Joint Surgery* [Br] **76**(1)113-117.

Versveld GA, Solomon A(1982). A diagnostic approach to tuberculosis of bones and joints. *Journal of the Bone & Joint Surgery* [Br] **64**(4)446-449.

Dave PK (2001). Osteoarticular tuberculosis. In: Essentials of tuberculosis in children. Seth V, Kabra SK. New Delhi, Jaypee Bros **2** 174-189.

Shannon FB, Moore M, Houkom JA, Waecker NJ (1990). Multifocal cystic tuberculosis of bone. *Journal of the Bone and Joint Surgery* [Am] **72**(7)1089-1092.

Kher AS, Bharucha BA, Kamat JR, Kurkure P (1993). Cystic tuberculosis of bones. *Indian Pediatr* **30**(5)676-678.

Kothari DS, Rathi SP, Kher AS, Meswani B, Pote M (1999). Imaging diagnosis. *Indian Practitioner* **25** (2)122-123.

Rafiqi K, Yousri B, Arihi M, Bjitro C, Aboumaarouf M, El Andaloussi M (2013). Unusual locations of osteoarticular tuberculosis in children: A report of 12 cases. *Orthopaedics & Traumatology: Surgery & Research Journal* **99**(3)347-51.

Rasool MN(2013). Tuberculosis of the hip region in children. *The Journal of Bone & Joint Surgery* **95**(14) 74.