

## Case Report

### A Case Report of Hydatid Disease in Long Bone

H. Fanian MD\*, M. Karimian Marnani MD\*\*

#### ABSTRACT

Hydatid cyst, caused by echinococcus granulosa, can produce tissue cyst everywhere in body. Skeletal cystic lesion is rare especially in long bones like tibia and because of its unusual presentation, its diagnosis may easily be missed, unless be kept in mind.

**Keywords:** hydatid cyst, bone, tibia.

Hydatid cyst is caused by the larval stage of the canine tape worm *Echinococcus granulosa* which produces tissue cyst throughout the body<sup>1,2</sup>. The incidence of bone disease is extremely low as most larvae are trapped by the liver and lung upon release of embryo in to the portal blood stream<sup>1,3,4</sup>. Skeletal cystic echinococcosis is found in only 0.5-2 % of cases of echinococcosis<sup>5,6</sup>. Anatomoclinical changes are, however, peculiar to this localization<sup>3</sup>.

From anatomopathologic stand point, this localization marks the torpid, insidious progression of the parasite into the bone tissue, leading to a diffuse, extensive, invasive process; so From the clinical stand point where ever it is localized, its complete surgical eradication is rarely possible<sup>3</sup>.

We surprised by the latency of this affection; the patient is being treated at an advanced stage, when radiologic lesions are already extensive, and complications, especially in the spinal area, are severe<sup>3</sup>. Owing to the poor biologic findings, the dagnosis of osseous hydatidosis is still primarily based on roentgenographic findings<sup>3</sup>. Then plain radiography, CT scan, and MR Imaging are helpful in the diagnosing skeletal cystic echinococcosis<sup>6</sup>.

Accurate diagnosis may aided in some persons by eosinophilia (25 to 35% of cases) and positive result of complement fixation tests, intradermal injection

of hydatid fluid (casoni test), and indirect hemagglutination tests<sup>6,7,8</sup> over a long period of time.

Osseous foci may be manifested as pain and deformity, particularly in 30-60 years old age group. Hydatid disease of bone is rarely seen in childhood<sup>7</sup>.

On the basis of available reports, the vertebral column, the pelvis, the long bones, and the skull are most commonly involved<sup>7</sup>.

A review of literatures shows that osteohydatid disease implicates the spine in 35%, the pelvis in 21%, the femur in 16%, and the tibia in 10% of cases. The ribs, skull, scapula, humerous, and fibula harbour cysts have low incidence; between 2 and 6% of the total cases of bone hydatid disease<sup>1,9</sup>.

As no connective tissue barrier form in bone, daughter cysts extend into bone, infiltrating and replacing medulla, leading to the constraints of this external layer, the cysts progressively enlarge, filling the medullary cavity to a variable extent<sup>1</sup>. Growth in the direction of least resistance, in time, causes cortical destruction with extension of the cyst into surrounding soft tissues<sup>1</sup>.

The cysts might lie dormant for as many as 20 years<sup>1</sup>, as it is several years before pain and deformity become apparent. the disease presents in middle age, generally between the fourth and sixth decades<sup>1</sup>. This condition is rarely encountered in childhood<sup>1</sup>.

\* Assistant Professor, Department of Orthopedics, Isfahan University of Medical Sciences, Isfahan, Iran.

\*\* Resident, Department of Orthopedics, Isfahan University of Medical Sciences, Isfahan, Iran.

Correspondence to: Dr Karimian Marnani, Department Of Orthopedics, Isfahan University Of Medical Sciences, Alzahra General Hospital, Isfahan, Iran.

## Case Report

We present the case of a 38 years old man who complained from left leg pain for one month. during this period and at the first visit, he had a tumor like mass in middle third of his left leg that was painful and slightly warm but not erythematous. he had not any important event in his history and hadn't any important medical or surgical history. He had suffered from limping due to painful leg in that period of time.

In evaluation of patient, WBC, diff was normal. Simple X-ray of left leg showed advanced lytic destruction of bone, centered at proximal two thirds of left tibia. At bone scan that was performed, active tumoral bone lesion of proximal half of the left tibia without any other lesion throughout the skeleton was reported. MRI showed a non homogenous lesion in left tibia. After primary evaluation, we took a biopsy of the lesion and during biopsy we encountered with cystic lesion that contained fluid and small soft tissue particle that was infected. Specimens were sent for further examination, and after that, hydatid cyst diagnosis was confirmed. Then in second procedure, we did curettage of lesion and filled tibial bone defect with corticocancellous bone graft from ipsilateral iliac crest and contra lateral fibular graft.

He has followed after surgery up to now, he has not taken any medication, during this period. He is completely symptom free and he came back to his normal life. Figures 1-5 show preoperative XRay , MRI , bone scan, and bone lesion after excision and follow up X-Ray) .

## Discussion

Although the incidence of hydatid disease has decreased as a result of education and control measures, there are still foci of concern in South American and sporadic cases still occur in United States, Europe, the middle East, and Asia <sup>4</sup>. Echinococcus is endemic in Iran <sup>4</sup>.

Skeletal cystic disease occurs in the more highly vascularized areas of the bones<sup>6,8</sup>.

The vertebrae, long bone epiphyses, ilium, skull, and ribs are most frequently affected<sup>6,8</sup>. The hydatid cysts may lie dormant in the bone for as long as 40 years<sup>6</sup> and most skeletal hydatid cyst cases have been adults<sup>6</sup>. Skeletal cystic echinococcosis lesion may be single or multiple<sup>6</sup>.



**Figure 1.** preoperative X-ray of left leg.

As hydatid disease of bone remains asymptomatic over a long period, it is usually detected after a pathological fracture or secondary infection (like our case) or following the onset of compressive myelopathy in cases of vertebral lesions<sup>1</sup>.

The most common radiological manifestation of skeletal hydatid disease is a lucent expansile lesion with cortical thinning<sup>1,10</sup>.

The differential diagnosis of skeletal cystic echinococcosis includes other infectious lesions, (e.g. tuberculosis), fibrous dysplasia, tumors (simple bone cyst, aneurismal bone cyst, plasmocytoma, osteosarcoma, chondrosarcoma, chondromyxoid fibroma, lymphoma, giant cell tumors, brown tumor, metastases, etc.)<sup>6,7</sup>.

The diagnosis is difficult since the more easily recognized involvement of other organs is rare<sup>6</sup>.

Daughter cysts, calcification, and germinal membrane detachment, typical manifestations of cystic echinococcosis in parenchyma organs, are not usually observed in skeletal hydatid cysts. The definitive diagnosis can usually be made by histopathological examination<sup>6,8</sup>.

In conclusion, the preoperative differential diagnosis of skeletal cystic lesions should include

cystic echinococcosis, since the diagnosis may easily be missed, unless be kept in mind.



Figure 2. Preoperative MRI, coronal, axial, and sagittal views at different sections.



**Figure 3.** Preoperative Bone scan



**Figure 4** Photograph of Hydatid lesion of surgical procedure



**Figure 5.** Postoperative X-Ray which shows fibular graft in tibial medullary canal.

## References

1. Morris B, Madiwale C, Gary A, Chavhan G. Case report: Hydatid disease of bone. *Australasian Radiology*. 2002 Dec; 46(4): 431.
2. Rong SH, Nie ZO. Hydatid disease of bone. *Clin Radiology* 1985; 36: 301-5.
3. Zliti M, Ezzaouiak, Lebib M, Karray M, Kooli M, Mestiri M.
4. Hydatid cysts of bone: Diagnosis and treatment. *World journal of surgery*. 2001 jan; 25(1): 75-82.
5. Fazel F, Ghanbary H. Hydatid cyst of the orbit. *Journal of Isfahan Medical School*. 2002; 20(65): 11-12.
6. Merkle E, Schulte M, Vogel T. Musculoskeletal involvement in cystic echinococcosis. *Report of eight cases and review of the literature*. 1997; 168:1531.
7. Tüzün M, Hekimoglu B. CT findings in skeletal cystic echinococcosis. *Acta Radiology*. 2002 Sep; 43(5): 533.
8. Resnick D. Osteomyelitis, septic arthritis and soft tissue infection. IN: Resnick. *Diagnosis of bone and joint disorders: From saunders*, 2002: 2510-2626.
9. Gossios K, Kantoyannis D, Dascalogiannaki M, Gowrtsoyiannis N. Uncommon Location of hydatid disease, CT appearances. *Eur. Radiology*. 1997; 7:1303.
10. Tüzün M, Hekimolu B. Various Location of cystic and alveolar hydatid disease: CT appearances. *J comput Assist Tomogr*. 2001; 25:81-7.
11. Booz M. The value of plain films in hydatid disease of bone. *Clin Radiol*. 1993; 47: 265-8