Hydatid Disease of the Left Femur
A Case Report

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Abstract
Osseous hydatid disease is a rare but serious condition. Treatment is difficult because of the progressive course of the bone involvement and generally admitted algorithm about osseous hydatid disease. We report a six-year follow-up of a case with involvement of the left femur, treated with an unconnected surgical method and albendazole. In this patient a 1 cm segment of the cortex between the trochanteric region to supracondylar area of the left femur was removed. The medullary cavity of the left femur was irrigated for 5 minutes with 20% hypertaline solution, and removed without causing any damage. The medullary cavity was curettaged meticulously and irrigated for 6 minutes with 0.9% saline solution. The bone defect was filled with bone cement. Albendazole was administered during the postoperative period. At the sixth year postoperatively, the patient was pain free. All serological tests were normal. Radiologic evaluation showed no evidence of disease recurrence. Meticulous preoperative planning, excision of all the cysts, and an effective regimen of chemotherapy will reduce recurrence. Bone scintigraphy is an important diagnostic method during the follow-up period.

Hydatid cyst disease is a parasitic disease caused by a cestode known as echinococci. The genus Echinococcus includes three species; Echinococcus multilocularis, Echinococcus vogeli and Echinococcus granulosus. Echinococcus granulosus is the most common cause of hydatid disease in man.1,2 The definitive hosts are dogs, foxes, and other carnivores. The tape worms live in the small bowel of these hosts and infected ova are shed in the feces. When ingested by intermediate hosts such as man, sheep, or cattle, the larvae enter the portal circulation. The larvae eventually reach the liver, where of them most are trapped. Sometimes, larvae reach the lungs and other areas of the body and form cysts. The life cycle is completed when the definitive hosts consume infested viscera of the intermediate host.1,2

The primary hydatid disease of the bone, caused by Echinococcus granulosus is formed when the scoleces are localized in the bone, and it is seen in 1% to 2.4% of the cases.1,2 The strong structure of osseous tissue limits the growth of the hydatid cyst, which spreads along medullar and trabecular channels. The trabeculae are slowly resorbed due to pressure without any cortical extension. The cysts extend to surrounding soft tissues if the bone cortex is eroded. The disease affects long bones, vertebral column, pelvis, and costae in order from least to most affected region.1,2 In general, the patients present to clinicians with local pain, swelling, or a pathologic fracture.

Giant cell tumors, solitary bone cysts, aneurysmal bone cysts, fibrous dysplasia, bone metastasis, neurofibromatosis, and tuberculosis of the bone should be considered in the differential diagnosis of osseous hydatid disease.4 More recently, special diagnostic techniques
such as computed tomography (CT) and magnetic resonance imaging (MRI) have been used for the differential diagnosis and accurate assessment of the extent of the disease.\textsuperscript{2-6}\textsuperscript{8} We present the six-year follow-up of a case with involvement of the left femur, treated with an alternative surgical method and albendazole. We also used bone scintigraphy together with CT and MRI.

Case Report

A 17-years-old female patient was admitted to the outpatient clinic suffering from persistent pain of four months duration that she described as dull in nature and extending from the left knee to the hip. A biopsy had been previously performed at another hospital and the diagnosis was “hydatid cyst in the bone tissue.” The patient’s history and the physical examination were normal. The erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) was 80 mm/h (N: 0-15 mm/h), and was 48 mg/dl (N: 0-6 mg/dl), respectively. Other biochemical values were normal. The \textit{Echinococcus} latex agglutination test was 1/256 + titers (N: 0/128). Radiographs showed cystic lesions extending from the left femoral neck to the supracondylar area of the left femur. Small calcifications in the trochanteric region and sclerosis as well as destruction of the diaphysis was observed (Fig. 1). Large cystic lesions filling the medullary cavity caused destruction and expansions in the cortex, extending from the trochanteric region to distal of the diaphysis, as demonstrated on a CT axial section of the left femur. Coronal T1-weighted MRI showed a large bone and adjacent soft-tissue hypointensity extending from the left femoral neck to the entire diaphysis (Fig. 2). Scintigraphy demonstrated that the osteoblastic activity was increased from the left femoral neck to supracondylar area (Fig. 3).

At surgery, the left femur was reached through a lateral longitudinal incision from trochanteric region to the above-knee level. After retraction of the soft tissues with Hohman retractors, a 1 cm width of the cortex was removed between the trochanteric region to the supracondylar area of the left femur. The entire medullary cavity including the femoral neck was irrigated for 5 minutes with 20% hypersaline solution, and was removed atraumatically without causing any damage to the germinative membrane. The medullary cavity was then curetted meticulously and irrigated for 6 minutes with a 0.9% saline solution that had been warmed to 90° C. The defect in the left femur was filled with bone ce-

\textbf{Figure 1} Anteroposterior radiograph of the left femur showing cystic lesions extending from left femoral neck basis to the supracondylar area of the left femur. There are multiple calcifications on the trochanteric region and sclerosis and destructions along diaphysis.

\textbf{Figure 2} Coronal T1-weighted MRI of the left femur, showing a large bone and adjacent soft-tissue hypointensity extending from the left femoral neck to the whole diaphysis.
ment. Suction drains were removed 48 hours postoperatively.

Pathological examination revealed that the cuticular membrane had a lamellar, acellular, homogenous eosinophilic appearance. In addition, soft tissue pieces showing fibroblastic proliferation, inflammatory cells, and infiltrated giant cells were observed. The pathological diagnosis was “hydatid disease of the bone tissue.”

Postoperative treatment started with albendazole 10 mg/kg/day administered orally. Partial weightbearing was permitted at the end of second month. Full weightbearing was permitted at end of third month. At the end of the 18th month administration of albendazole administration was discontinued.

The physical examination performed six years postoperatively was normal. The patient was pain free. On control radiographs, a hypodense area approximately 1 cm thick (bone cement filling the defective area) was seen between the trochanteric region and supracondylar area of the left femur. Destruction was not observed although expansion and thinning were seen in some areas of the cortex (Fig. 4). Increased osteoblastic activity and no recurrence of disease can be seen on the bone scintigraphy that was also performed at this six-year follow-up (Fig. 5). Control ESR 10 mm/h and Echinococcus...
latex agglutination test was 1/10 titers.

Discussion

The radiological signs of the osseous hydatid disease include lucent lesions in the bone that are associated with bone expansion and thinning of the cortex. In patients with these signs, soft tissue calcifications are highly suggestive of hydatid disease. CT and MRI are important in determining the accurate extent of the bone and soft tissue abnormalities.

The literature on bone scintigraphy was reviewed but no report concerning osseous hydatid disease was found. However, there are some reports about hydatid disease in the liver and the chest. These reports emphasize that in a great percentage of cases scintigraphy shows accurate localization, dimension of the cysts, and assists the surgeon by establishing the most appropriate surgical approach to the cyst thus reducing the danger of spilling its content. According to us, the bone scintigraphy is also the most significant method for the purposes of differential diagnosis; additionally it delineates the accurate extent of the bone lesions and is also useful in assessing recurrence during the postoperative period. Scintigraphy should be employed for the differential diagnosis, preoperative planning, and in the follow-up period of the osseous hydatid disease.

There is no generally accepted treatment algorithm for osseous hydatid disease. Cases of osseous hydatid disease are rare and treatment methods are generally unsuccessful. The literature offers various treatment approaches. Until recently, the basic treatment has been surgical excision or resection. Unfortunately, the results have been discouraging. Surgical excision and curettage are the basic approach for osseous hydatid disease. Only, macroscopic cysts can be extracted with surgical excision and curettage. The surgical area might be irrigated with chemical agents in an attempt to kill scoleces. According to us, the bone scintigraphy is also the most significant method for the purposes of differential diagnosis; additionally it delineates the accurate extent of the bone lesions and is also useful in assessing recurrence during the postoperative period. Scintigraphy should be employed for the differential diagnosis, preoperative planning, and in the follow-up period of the osseous hydatid disease.

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The combination of chemotherapy and surgical treatment has been found to be more efficient than surgical treatment alone. Mebendazole, albendazole, and antihelmintic drugs are used for chemotherapy. Albendazole has been found to be better absorbed than mebendazole and exhibits superior efficacy against helminths. Bonifacino and colleagues administered oral albendazole (15 mg/kg/day) in 28-day cycles. The efficacy of chemotherapy was assessed by serological tests for circulating antigen, immunocomplexes and specific antibodies, and by radiography and CT scans. They concluded that treatment with albendazole is effective, but one cycle should be given before operation and six or more courses afterward.

Uzel and associates stated that *Echinococcus granulosus* infestation of the bone is a progressive tumor, similar to a bone tumor. They emphasized that bone cement and local heating applied after curettage are an effective method for treating this disease. They applied this method in the treatment of two cases—one case was cured after 12 months the other after 30 months. However, these investigators saw the recurrence of fistula formation in 6 cases in which treatment with chemical agents and curettage had been applied.

We also think that for achieving successful results in the treatment of the osseous hydatid disease, this disorder should be considered as a tumor of progressive nature. The treatment algorithm must take into consideration this thesis. The first step in the treatment should be meticulous preoperative planning that includes special diagnostic techniques such as CT, MRI, and bone scintigraphy. In this way, the extent of the bone lesions can be determined precisely. This approach reduces the possibility of the recurrence. The second step is an aggressive curettage of the affected bone after irrigation with 20% hyper-saturated saline solution at least 5 minutes and then irrigation for at least 5 minutes with 0.9% saline solution heated to 90°C. Then, after irrigation, bone cement is applied to the defect, which has the additional beneficial effect of heating and toxic polymerization. The third and last step is an effective chemotherapy program for at least 12 months. It should not been forgotten that osseous hydatid disease can recur any time. Therefore, patients without any symptoms should be followed for the long-term and serological tests, radiographs, and bone scintigraphy should be used periodically to assure that the disease has not recurred.

References


