Bone Hydatidosis of Femur: A Case Report

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Abstract

Background : Hydatidosis is an infection caused by larval form of Echinococcus Species. It primarily involves the liver and lungs. Primary skeletal involvement is rare. The location of hydatid cysts in the femur is seldom described in the medical literature, and its diagnosis is challenging and often presenting with a pathologic fracture simulating benign bone cystic lesion.

Keywords: Anti-helminthics, curettage, hydatid disease, long bones

Introduction

Echinococcosis is an endemic zoonosis caused by adult or larval stages of cestodes belonging to the genus Echinococcus. Hydatid disease or Hydatidosis is characterized by long-term growth of metacestode (hydatid) cysts in the intermediate host. The two major species of medical and public health importance are Echinococcusgranulosus and E. multilocularis, which cause cystic echinococcosis (CE) and alveolar echinococcosis (AE), respectively. The disease is chronic and cyst can localize in different organs, most commonly in the liver(50-77%), lungs(8.5-43%), brain(3%) and even in skeletal muscles but osseous hydatidosis is a rare occurrence of hydatid disease, with an incidence of about 0.5-4%. Owing to the poor biologic findings, the diagnosis of osseous hydatidosis is still primarily based on roentgenographic findings. Sometimes, however, the diagnosis is established only after surgery.

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Case Report

We report the case of a 48 year-old female who presented to the OPD of Department of Orthopedics in Vydehi Institute of Medical Sciences with complaints of swelling and a discharging wound over proximal anterolateral surface of right thigh since 6 months which was associated with pain over the entire right thigh. On further evaluation patient a gives history of diffuse pain in the right lower limb since the past 8 years which has progressed to inability of the patient to walk or bear weight over the affected limb. On physical examination, there was diffuse swelling of right thigh, over the antero-lateral aspect with a discharging sinus at the summit of the swelling. The swelling roughly measured about 20 cmx20 cm with the sinus measuring about 1 cmx1 cm. It was cystic in consistency and was associated with intermittent purulent discharge. Routine laboratory tests were normal. Clinical picture of an abscess was kept in mind and the patient was taken to the department of radiodiagnosis for ultrasonography of the right thigh. The ultrasound scan showed a large hyperechoic collection with septations and a few solid areas, circumferentially involving the right thigh which was seen extending below up to the distal 1/3rd of right thigh, with the predominantly cystic component in the antero-lateral aspect and the solid component in the medial aspect, and underlying femur showed irregularity - features likely of osteomyelitis. X-ray findings of the right Femur showed thinned out and irregular cortices, cortical sclerosis and an expansile lytic lesion distributed primarily over the proximal 2/3rd of the femur including the acetabulum with resorption of the head and multiple pathological fractures. A provisional diagnosis of chronic osteomyelitis with bone destruction and the abscess was made and the patient was taken up for emergency incision and drainage. Incision was made over the swelling on the antero-lateral aspect of right thigh to find multiple cysts. Cysts were collected en-masse and some cysts were ruptured and the sac excised. Muscles were found to be hypoplastic with reduced vascularity and extensive fibrosis with loss of compartmental demarcation. Hydatid sand was seen as viscous fluid from the medullary canal. After thorough debridement and wash, closure was done with successive regular dressings. After consulting the microbiologist, chemotherapy in the form of oral albendazole and praziquantel was started. In view of the

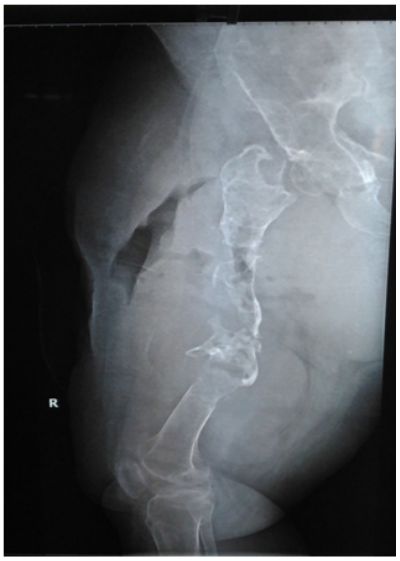

Fig. 1:Plain radiograph of right Femur showing Osteolytic lesion with pathological fracture, thinned out cortices with absence of femoral head.

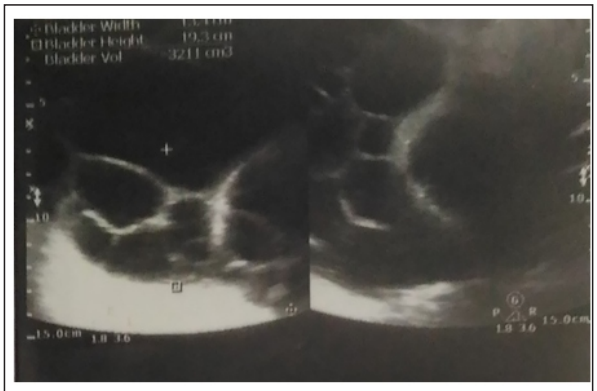

Fig. 2:USG showing large heteroechoic with septations and some solid areas.

severely damaged musculo-skeletal compartments, arthroplasty or fixation of the femur could not be planned. Consequently, a second surgery was planned due to persistent discharge from the lateral surgical site with the postoperative magnetic resonance imaging (MRI) in view, which showed multiple large cysts in the antero-medial compartment of the right thigh which was not visualized intraoperatively. After 3 months of therapy with anti-helminthics, with an incision from the medial side of thigh, a thorough debridement was done excising as many cysts as possible from the antero-medial compartment and superiorly up to the inguinal ligament. After irrigation with saline, wound closure was done in layers with suction drains secured within the medullary cavity. No anaphylactic reactions were noted during the surgery. Post operatively anti-helminthics were continued with regular dressings.

Patient was presented with two possible modes of management in the form of:

1. Stage 1 - Resection of upper 2/3rd femur with placement of antibiotic cement spacer/followed by Stage 2 - tumor megaprosthesis or
2. Hind quarter amputation of the affected limb followed by rehabilitation.

The patient was asked to follow-up at regular intervals and discharged.

Discussion

Hydatidosis of the bone normally involves the spine and pelvis. The disease is mostly secondary, usually due to hepatic or pulmonary hydatidosis. It might in some cases appear as an isolated primary disease [1]. Bone hydatidosis can be suspected from radiographs, computed tomography, and MRI. Diagnostic serological tests for hydatid bone disease (ie, counter-immunoelectrophoresis, Casoni, gold antibody, and indirect hemagglutination) mainly revolve around the detection of antigen-5 and antigen-B in the hydatid fluidor serum antibody. ELISA for hydatidosis has the highest sensitivity and specificity and can determine the response to treatment during medical therapy [2]. Most of the times, hydatid cysts remain asymptomatic for a long duration and are detected only after a pathologic fracture or a secondary infection [3]. Radiographic findings in bone hydatidosis include intramedullary unilocular, bilocular, and often multiloculated cystic expansile lesions with surrounding sclerosis in a honeycomb pattern associated with cortical thinning. Soft tissue calcification surrounding these lytic lesions is occasionally present. Typical MRI findings include a high-signal appearance of a rose or wheel-shaped lesion in T2-weighted images due to the presence of spaces orseptation in the daughter cyst [4]. Diagnosing bone hydatidosis is difficult considering its asymptomatic mode of presentation and rarity of the lesions, and also the absence of typical radiographic features to

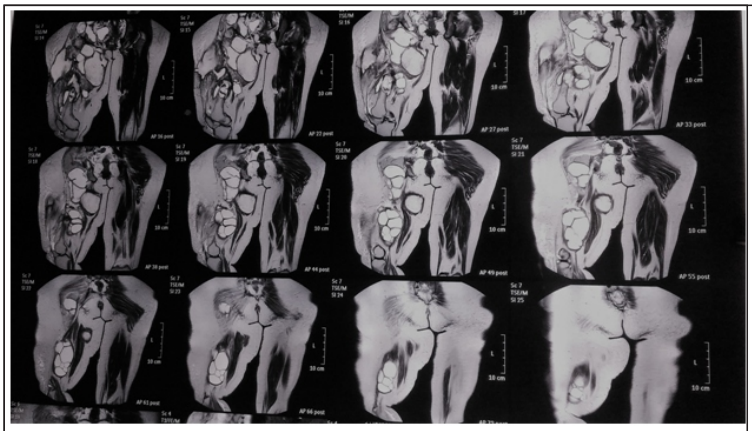

Fig. 3: MRI of the right thigh showing multiple intramedullary and extra-osseous cysts.

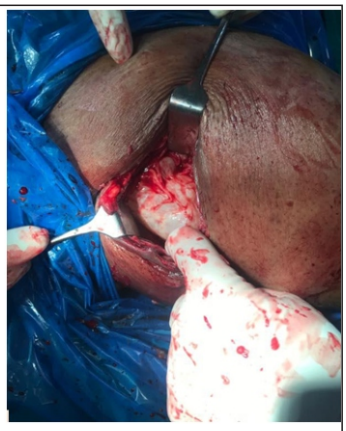

Fig. 4:Femur exposed through lateral approach

Fig. 5:Numerous cysts evacuated of varying sizes.

definitively diagnose bone hydatidosis [5]. A combined surgical and chemotherapeutical intervention is the most effective mode of management of bone hydatidosis [6]. Surgical management comprises radical resection of the diseased segment along with a wide margin of soft tissues. Residual soft tissue may be a source for recurrence, hence thorough debridement with cyst excision along with diseased tissue with wide margin is recommended. Possibility of anaphylactic shock should be considered in case of intra-operative cyst ruptures or during debridement [7]. Deaths have been reported during intra-operative cyst rupture or spillage and subsequent anaphylactic shock. Various chemotherapeutic agents have been used for the treatment of human echinococcosis. Benzimidazole compounds, mebendazole and albendazole, have been used more frequently than others for the treatment of osseous and extraosseoushydatid disease [8]. Albendazole has better absorption and achieves higher concentration of its active metabolite in cysts and the blood compared to mebendazole and is considered the treatment of choice in human hydatid disease. Local scolicidal agents have been also tried either intraoperatively or postoperatively through irrigation with local drain intact to attempt local eradication of the disease [9,10].The commonly used scolicidal agents are 3% hypertonic saline, 10% formalin, hydrogen peroxide, chlorhexidine, 80% alcohol, povidone-iodine-alcohol solution or 0.5% silver nitrate. These agents help eradicate daughter cysts but not necessarily local recurrence of the disease [11]. Radiotherapy has not proven to be effective in the treatment of Hydatid disease of the bone [12].

Conclusion

Bone Hydatidosis is a rare condition which needs to be considered as a differential diagnosis in cases of suspected osteomyelitis. Long term follow-up is needed for better understanding of treatment. Multi-disciplinary approach is required in treating bone hydatidosis.

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