



An unusual cause of knee pain in a young patient; hydatid disease of femur

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ARTICLE INFO

Article history:

Received 10 April 2012

Received in revised form 3 May 2012

Accepted 13 May 2012

Available online xxx

Keywords:

Osseous

Hydatid

Disease

ABSTRACT

INTRODUCTION: Osseous hydatid disease is a rare pathology and its differential diagnosis can be difficult.

PRESENTATION OF CASE: This article presents clinical and radiological findings of a femoral primary hydatid disease in a 23 year-old patient admitted with knee pain.

DISCUSSION: Osseous hydatid in femur is a rare entity. Curettage is one of the surgical options with high risk of anaphylaxis and implantation. Radical resection of involved segment is the preferred treatment method in the current literature. However, patient preference can be different.

CONCLUSION: Although late stages of this disease need be treated with amputation or disarticulation, in the early stages it can be treated with curettage and bone cementing. Our case with early diagnosis of hydatid disease of distal femur was treated successfully with curettage and bone cementing.

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1. Introduction

Osseous hydatid disease is a rare entity in which early diagnosis can be difficult and treatment can be challenging. In early stages of osseous hydatid, pain can be the only symptom.^{1–3}

Knee pain is one of the most frequent admission symptoms to orthopedic outpatient clinic. Knee pain can be due to traumatic or non-traumatic pathologies; such as overuse injuries, septic arthritis, inflammatory conditions, Charcot joint, osteochondral lesions, and neoplasm.⁴

In this case report, a rarely seen disease osseous hydatid cyst of femur in a 23 year-old patient with the only symptom of knee pain, has been presented in the light of the literature.

2. Case presentation

A twenty three year old female, presented to our outpatient clinic with complaints of right knee pain. She had had these symptoms for 8 months. Physical examination of her right knee revealed 30 degrees of flexion contracture and moderate swelling. There was no marked tenderness at peri-articular femur and tibia. X-ray examination revealed osteolytic lesion in the distal diaphysis of right femur (Fig. 1). Complete blood count was in normal ranges. Erythrocyte sedimentation rate was 43 mm/h. Systemic examination of the patient was normal.

There was no history of contact with potentially contaminated food or drinks. The patient experienced dog bite ten years ago from her right leg. We did not suspect hydatid disease at that time because of ingestion of ova bite definitive host is needed for infection. Our first pre diagnosis was osteosarcoma of femur with her previous clinical laboratory and radiographical findings. A fine needle aspiration biopsy was performed to confirm the diagnosis. Computerized tomography (CT) and magnetic resonance imaging (MRI) of distal femur were also performed in order to evaluate the extension of the disease in her right femur (Fig. 2). Additionally, head CT, abdominal CT, thoracic CT, and bone scan were obtained to evaluate the potential extension sites of the disease. CT scans were in normal range, bone scan revealed increased activity in her right femur diaphysis.

Histological examination of the biopsy material documented the characteristic findings of hydatid disease. Additionally, serological tests for hydatid disease that were performed at the time of presentation documented as positive. Oral Albendazole treatment was started 15 mg/kg for 20 days as the neo-adjuvant treatment. After neo-adjuvant treatment, prominent decrease of knee pain was obtained. Range of her right knee motion did not improve after Albendazole treatment. In the operation, marked increase of periosteal thickness in the diaphysis of right femur was observed (Fig. 3a). Green-brown purulent fluid was detected during curettage of the femur (Fig. 3b). Surgical treatment included curettage and cementing of femur (Fig. 3c). Postoperative X-ray examination revealed fully filling of the lesion with bone cement (Fig. 4). A cycle of Albendazole adjuvant therapy was started just after the surgery with the same dose. The duration of the adjuvant therapy was performed to be 20 days after consulting with the department of infectious diseases. We decided the duration of medical treatment with the serological tests.

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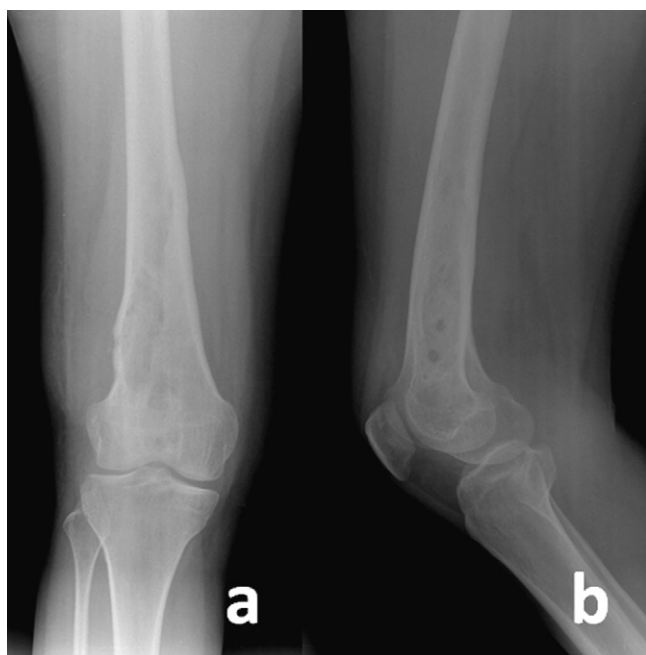


Fig. 1. (a) Anteroposterior and (b) lateral preoperative radiographic view of distal right femur.

3. Discussion

Hydatid disease is parasitic zoonosis caused by larval form *Echinococcus Granulosus*.² Even the incidence of systemic involvement is 59–75% in the liver, 27% in the lungs, 3% in the kidneys, and 1–2% in the brain.³ In our patient, no other involvement was determined with bone scan, head CT, abdominal CT, and thoracic CT.

Chaussier firstly reported bone hydatid disease in 1807.¹ The incidence of osseous hydatid disease is reported to be 0.5–4%.⁵ Spine is the most common skeletal involvement site (35–50%) followed by pelvis, tibia, femur, humerus, skull, and ribs.^{6–8} The osseous cysts remain asymptomatic and radiological changes are non-specific. One of the main features of bone hydatidosis is slow growth due to rigid structure of cortical bone.¹ Thus, it can be asymptomatic for decades. The diagnosis is often established in the late stages of parasitic extension.⁹ Osseous hydatid disease does not affect survival however it is not easy to eradicate. The severity of

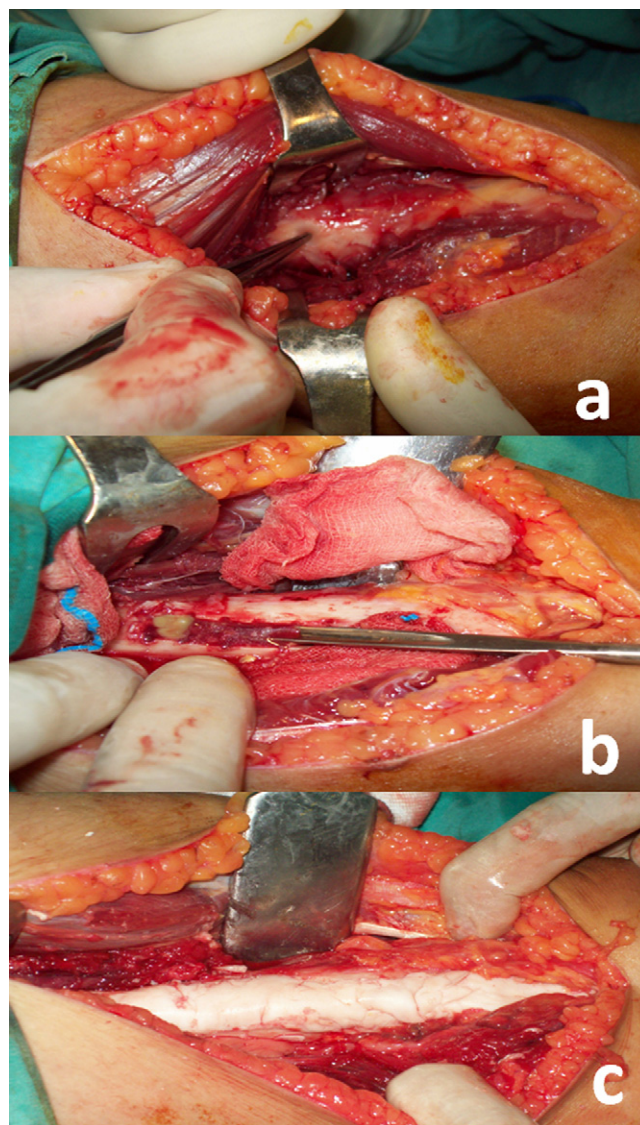


Fig. 3. Intraoperative pictures. (a) Cortical thinning and weakening of distal femur. (b) Green-brown purulent material in the distal femur. (c) Bone cementing after curettage of the lesion.

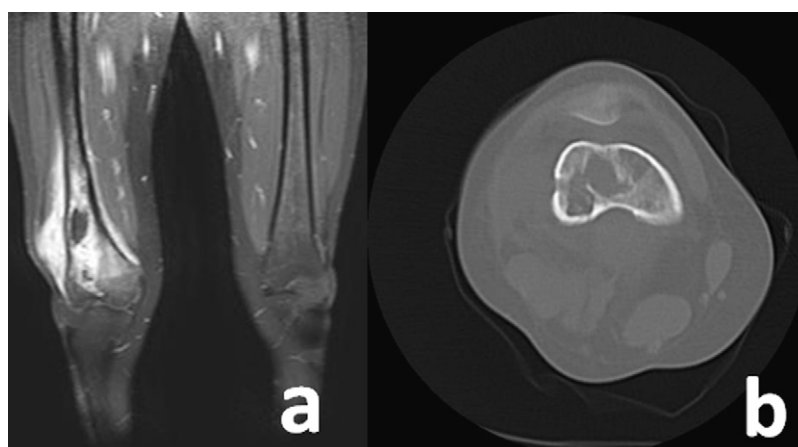


Fig. 2. Preoperative (a) MRI and (b) CT scan of osseous hydatid disease.

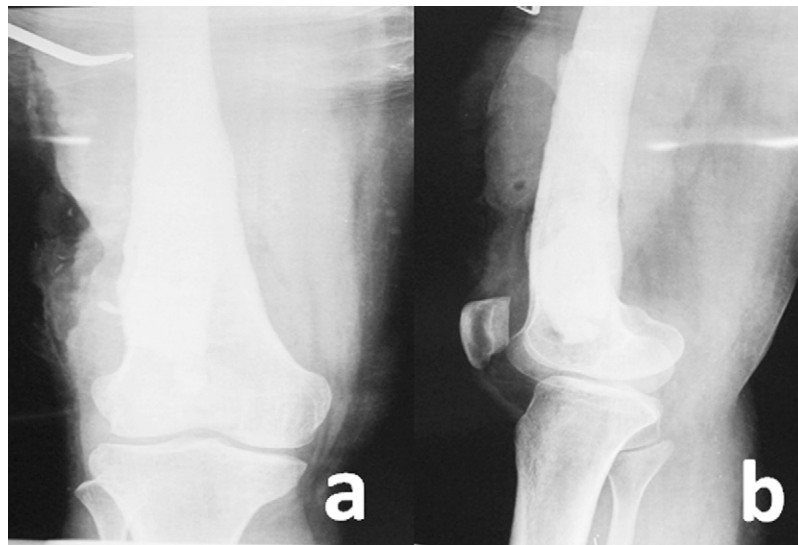


Fig. 4. (a) Anteroposterior and (b) lateral postoperative radiographic view of distal right femur.

prognosis led to name osseous hydatid disease as 'white cancer'.¹⁰ Our patient represented with right knee pain, and osseous lytic lesions were detected in her X-ray examination (Fig. 1). Additionally, CT and MRI of distal femur were performed not only for differential diagnosis but also for evaluating extension of the disease (Fig. 2).

Fecal oral transmission should be presented between the definitive and intermediate host. Embryos should pass hepatic and pulmonary filters to form cyst in skeletal muscles, bones, brain, and other tissues.¹¹ In this case contrary to current literature direct contamination with dog bite was detected.

Markonis et al. reported the mean age of diagnosis as 52 years and more frequent incidence in men.¹² Our patient was 23 year-old female who can be assumed early diagnosis in the light of current literature.

Macroscopically there is no clear delineation between healthy and infected bone in osseous hydatid disease. Small vesicles of various sizes, which rarely exceed 2 cm, infiltrate bone in this pathology.¹¹ Cortical infestation may cause involvement of adjacent soft tissue.¹ In our patient, we observed soft tissue involvement with MRI (Fig. 2a) and we detected some small vesicles with green-brown fluid material in the operation (Fig. 3b).

Presenting symptoms are usually non-specific like pain and swelling. Patients may present with symptoms due to complications, such as fracture, fistulization. These frequent non-specific clinical presentation causes difficulty in differential diagnosis of bone hydatidosis especially from bone tumors, myelomatosis, lymphoma, aneurysmal bone cyst, brown tumor of hyperparathyroidism, and chronic osteomyelitis.^{1,11}

Standard X-rays usually reveal single or multiple expansile osteolytic lesions with cortical thinning.¹³ Osteosclerosis may be seen in later stages and periosteal reaction may be a sign of pathological fracture.¹⁴ Metaphysis is the initial lesion site in long bones. At later stages diaphyseal involvement is seen with spared general appearance of bone and articular spaces.⁸ CT reveals non-specific changes but it is valuable in defining extension and precise location of disease and also possible fractures.⁷ MRI is most helpful radiologic tool for bone hydatidosis¹⁵ where it may also document adjacent soft tissue involvement.¹⁶ We performed both CT and MRI for accurate diagnosis preoperatively (Fig. 2).

Serological tests are useful to confirm diagnosis but some of the patients do not show detectable immune response.¹⁷ Bone

hydatosis causes higher level of serological response due to lack of pericyst.¹¹ Serological tests are also useful in screening and follow-up. Complete blood count is usually in normal range where eosinophilia is not always present.¹¹ Our patient's complete blood count was in normal ranges without eosinophilia. And also, positive antibody levels of serological tests were obtained.

Until 1980s surgical treatment was the only option for hydatidosis. However, medical treatment with Benzimidazole compounds, treatment with cyst puncture, aspiration, injection of chemicals, and re-aspiration have been suggested as other options.¹⁸ When possible, surgery remains the best treatment method for the immediate and complete cure of the disease. Curettage as one of the surgical options has high risk of anaphylaxis and implantation.¹⁹ Radical resection of involved segment just as in the treatment of malignant lesions is recently recommended surgical approach.⁸ Contraindications for surgery are refusal of the patient, pregnancy, and anesthesia related contraindications. Operative mortality is reported between 0.5 and 4%.¹⁸ In our case, we suggested hip disarticulation but patient refused this and also she refused transfemoral amputation. Therefore, we performed curettage and bone cementing.

Adjuvant medical treatment may be given preoperatively and postoperatively to control activity of the disease, and to avoid systemic spread and recurrence.¹¹ Benzimidazole compounds provide cure for 1/3 of patients, and significant disease activity regression in 30–50% of patients.²⁰ Both Albendazole (10–15 mg/kg/day) and Mebendazole (40 mg/kg/day) demonstrated efficacy. Albendazole has superior results because of its pharmacokinetic profile.¹⁸

Serological tests can be used to monitor treatment. Objective response to treatment is best assessed with evaluation of the lesion site with CT or MRI at regular intervals. Since the appearance of recurrence is variable monitoring should continue for at least 3 years.¹¹ We used serological tests and MRI for follow-up and we decided to end the medical treatment due to serological tests.

4. Conclusion

Bone hydatid disease is a very rare entity. Its diagnosis can be interfered with neoplasms, osteomyelitis or other bone diseases. In early diagnosis it can be successfully treated with curettage and bone cementing.

Conflict of interest statement

None.

Funding

None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contributions

Ersin Kuyucu (study design).
 Mehmet Erdil (data collecting).
 Ali Murat Dulgeroglu (writing).
 Figen Kocyigit (data collecting and analysis).
 Arslan Bora (figure preparation).

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