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Case Report

Hydatid Cyst Located in Knee Joint and Tibia: Case Report

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Abstract

Hydatid cyst is a parasitic infection caused by the parasite known as *Echinococcus granulosus* and mostly enjoying localization in liver. Localization in bone or soft tissues is rarely seen. In our study, the case of a 71 years old male patient diagnosed with hydatid cyst of bone localized at left knee joint is presented and the literature is reviewed.

Keywords

Bone; *Echinococcus granulosus*; Hydatid cyst; Tibia

Introduction

Echinococcosis (hydatid cyst) is the disease caused by larval forms of *Echinococcus* [1]. There are 12 different species, and among all, *Echinococcus granulosus*, *Echinococcus multilocularis* (alveolar hydatid cyst), *Echinococcus vogeli* (polycystic echinococcus) and *Echinococcus oligarthrus* cause disease in humans [2,3]. Parasites are more common in sheep and cattle breeding countries.

Main hosts of hydatid cyst causing *Echinococcus granulosus* are animals such as dogs, wolves and foxes, while intermediate hosts are sheep, goats and cattle [2].

After the parasite eggs are taken orally, they reach the liver through the bloodstream from the gastrointestinal tract and cause disease in other organs through the systemic circulation. Internal organ involvement, especially liver (70%) and lungs (10-15%), is seen [3]. As stated in the literature, 1-2% primary bone and 1-4% soft tissue placement is seen [4-6].

Although Hippocrates, Aretaeus and Galen had used several terms about hydatid cyst in history, Pallas was the first to identify lesions similar to hydatid cyst in humans in 1766 [1]. In this study; we aimed to present a rare bone and joint involvement hydatid cyst case accompanied with the literature.

Case Report

A seventy two year old male patient presented to our clinic for a recurrent hydatid cyst found on biopsy materials accompanied by lingering leakage and pain, and that he had been operated on 4 times 6 months apart in different clinics due to reccurrence of symptoms from the first hydatid cyst operation which was 4 years ago. On first examination, leaking wound was present at the proximal of incision and near incisional scar tissue of around 15 cm, on left anterolateral tibia. Besides the temperature rise around the knee joint rash, swelling and tenderness was detected. Left knee movements were painful and limited. Patient could stand up by support but was not able to fully step on his left leg. In the examination of patient's blood, the values were as follows: sedimentation 60 mm/h (0-20), C-Reactive Protein (CRP) 20 g/L (0-6), eosinophil 6.2% (0.9-2.9).

In direct radiography (Figure 1a,b), knee Computed Tomography (CT) (Figure 2a-c), and knee MRI evaluations; intraosseous cystic locular regions were present at upper end of tibia related to joint (Figure 3a,b). After all abdominal Ultrasonography (USG), abdominal CT and lung CT examinations, as no involvement was detected of other organs

including liver and lungs, primarily bone and soft tissue hydatid cyst was diagnosed under spinal anesthesia after entering left knee anteromedial distal tibia from approximately 20 cm of longitudinal incision, tibia was resected up to15 cm and necrotic tissues were removed, and then, after washing the knee joint and the area, antibiotic spacer was inserted and the structures were anatomically closed (Figure 4a,b). Reconstruction with resection tumor prosthesis was planned (Figure 5a,b).

Pathological report of the resected material was compatible with hydatid cyst (Figure 6a,b). In the third month controls after surgery sedimentation, CRP and eosinophilia were reported as 12 mm/h, 5.3 g/L, and 5.6%, respectively. After that prosthetic reconstruction was performed.

Figure1a,b: Patient's AP and lateral X-ray graphs; in the area indicated by the arrow intraosseous loculated areas and bone grafts from previous operations are seen.

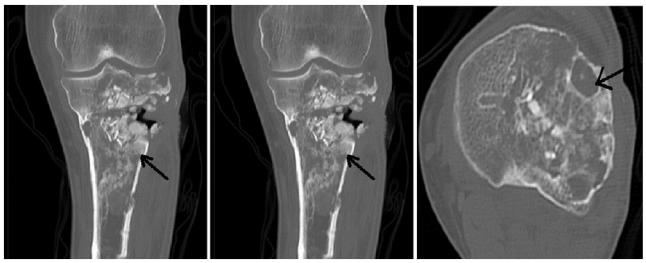

Figure2a-c: Preoperative Computerized Tomography sections of the patient.

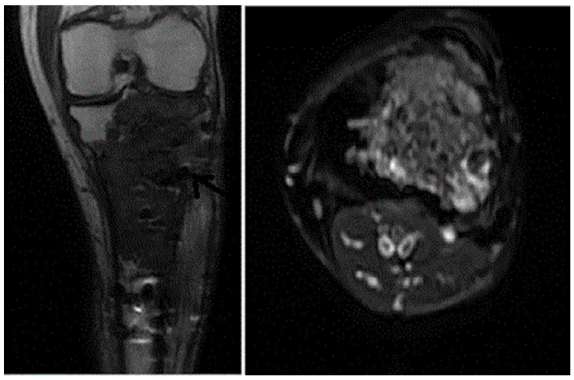

Figure 3a,b: Preoperative MRI sections of the patient.

Discussion

Hydatid cyst disease caused by *E. granulosus* is a common zoonosis [6]. Orally introduced larvae penetrate intestinal epithelium, transported to the target organs by blood and parasite forms hydatid cysts in these regions. These cysts are mostly seen in liver and lungs; involvement of kidney, spleen and bones is rare. Bone involvement is around 1-2%. It is most frequently seen in vertebrate (30-50%) and pelvic (15%) bones [6].

They are traced by long bone involvements such as femur, tibia and humerus [7]. Progress of the cyst in bones is slow. Damage of hydatid cyst in the human body is basically by mechanical means. However cyst formation in vital organs

Figure 4a,b: The graphs showing the region filled with antibiotic spacer after proximal tibia resection.

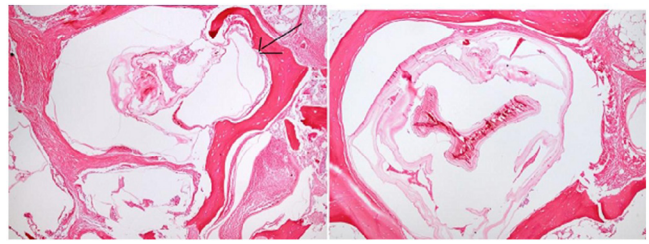

Figure 5: Specific Echinococcal areas occurred in pathological specimens of patient's resected materials.

a: Intramedullary localized hydatid cyst cuticular membrane pushing bone spicules is seen (HE, X100);

b: Intramedullary localized and concentric layering displaying cuticular membrane is seen (HE, X200).

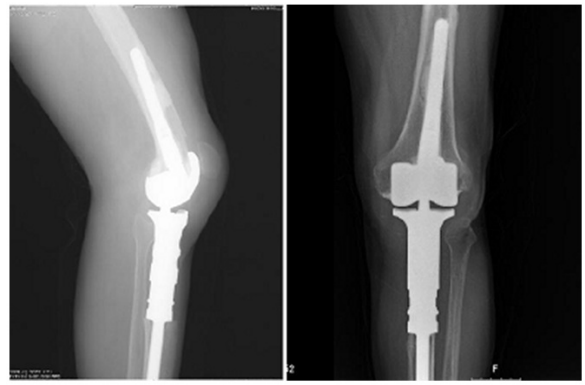

Figure 6a,b: Graphs of the knee joint reconstructed with tumor resection prosthetics after final surgery.

prevents the organ's functions and may cause damages resulting in death.

Unilocular hydatid cysts in the bone are called osseous [8]. Tuberculosis, simple bone cyst, fibrous dysplasia, malignant fibrous histiocytoma, sarcoma and metastasis should be considered in the differential diagnosis our case, in the examinations by external centers, was also assessed as simple bone cyst and operated on for cyst curettage four times.

The symptoms of osseous hydatid can be pain, swelling, deformity, pathological fractures and secondary infections. *Echinococcal* joint disease usually develops secondary to spread from adjacent bone [9]. There was spread to the joint in our case too.

Eosinophilia is a frequent indication in laboratory examination but sometimes it can be absent, tests such as ELISA, Western Blot, IHA, polymerase chain reaction are also useful in serological diagnosis [10]. While the serological sensitivity is 80-100% and specificity is 88-96% in liver involvement, it's reported to have 50-56% sensitivity in lung involvement and 25-56% sensitivity in other organ involvements [10]. Besides it is stated that *E. granulosus*-specific IgE levels are dependent on location of the cysts. While the sensitivity for liver hydatid cyst is 92%, it has been found to be 61.5% for bone involvement [10]. Also, positive IHA result is meaningful for diagnosis, but even the result is negative it still doesn't rule out the disease.

Radiologically wall thickening, calcification, daughter vesicles and germinating membrane separated from the wall are hydatid cyst specific findings. Because the spongoid bone is involved first, lucent lesions are seen with bone expansion and the cortex becomes thinner [8].

These findings can be evaluated more specifically by especially MRI [7,8]. In our case, the diagnosis was confirmed in the laboratory and radiological examinations beside the previous biopsy.

There is no generally accepted protocol for the treatment of bone involved hydatid cyst. This is because; number of cases is small and the treatment results are poor. In the literature, use of various treatment methods is seen [11,12]. Basically, resection has an important place in the treatment of bone hydatid cyst. After excision, irrigation of cyst pouch is performed with various chemical agents, but these kinds of treatments are palliative and relapses can generally be seen afterwards.

In this disease, often associated with relapse, good results can be obtained by explicit surgical resection and medical treatment [11,13]. In cases that the surgical total excision cannot be applied, Albendazole therapy with surgical treatment can yield more successful results.

In our own case, as the cyst was considered as infected, and it was settled at the upper end of the tibia and spread into the knee, excision and knee reconstruction were planned. Albendazole therapy was not applied as no other organ involvement was present.

The patient with receding laboratory values din not delay his follow-up checks did not encounter any recurrence. We believe that, tibial resection and reconstruction procedures in cases of hydatid cyst, is a good option in its current form.

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