

## Primary Intraosseous Hydatid Cyst of Femur

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**Introduction:** Echinococcosis is a parasitic and zoonotic disease of animals and humans. The cause is *Echinococcus granulosus* and occasionally, *Echinococcus multilocularis*. Hydatid cysts are mostly seen in the liver and lungs, although almost all organs and systems can be involvement. Hydatid cysts seen with bone involvement comprise approximately 3% of all hydatid cysts. Even if a long period of survey is possible, it is still difficult to eradicate the disease and effect a cure.

**Case Presentation:** In this study, an evaluation was made of a patient referred at Yozgat State Hospital Orthopedics and Traumatology Polyclinic with complaints of pain in her left thigh close to the knee. After examinations of plain radiographs, computerized tomography, magnetic resonance images, and blood parameters, a diagnosis was made of left femoral intramedullary hydatid cyst from excised intraoperative material. Throughout a 6-month follow-up period, there was no recurrence and functional results were good.

**Conclusions:** Based on this report (of a patient presented with an intramedullary cyst in the long bones), the primary bone hydatid cyst disease should be kept in mind and be investigated in the differential diagnosis.

**Keywords:** *Echinococcus granulosus*; Femur; Cyst; Hydatid

### 1. Introduction

Echinococcosis is a parasitic and zoonotic disease of animals and humans. The causative agent is *E. granulosus* and occasionally, *E. multilocularis*. Hydatid cysts are most often seen in the liver and lungs, although there can be involvement of almost all organs and systems. Hydatid cysts seen with bone involvement comprise approximately 3% of all hydatid cysts (1). Even if a long period of survey is possible, it is still difficult to eradicate the disease and effect a cure (2, 3). In this report of a patient presented with an intramedullary cyst in the long bones, it was emphasized that primary bone hydatid cyst disease should be kept in mind and be investigated in the differential diagnosis.

### 2. Case Presentation

A 43-year-old female referred at our polyclinic with complaints of pain in her distal area of the left thigh, which had been ongoing for approximately 6 months. She was admitted to a state hospital in Yozgat, Turkey in February 2014. The patient had a history of a dog bite in the lateral distal left thigh 15 years ago. On physical examination, there was pain on deep palpation of the distal area of the left thigh. No pathological lymph node was determined on palpation of surface lymph nodes.

In the laboratory tests, apart from erythrocyte sedimentation rate of 67 and high C-reactive protein (5.7 mg/L), no pathology was determined in routine haemogram and bio-

chemical tests. In the radiographic examination, there was a left femur distal diaphyseal-metaphyseal radio dense area.

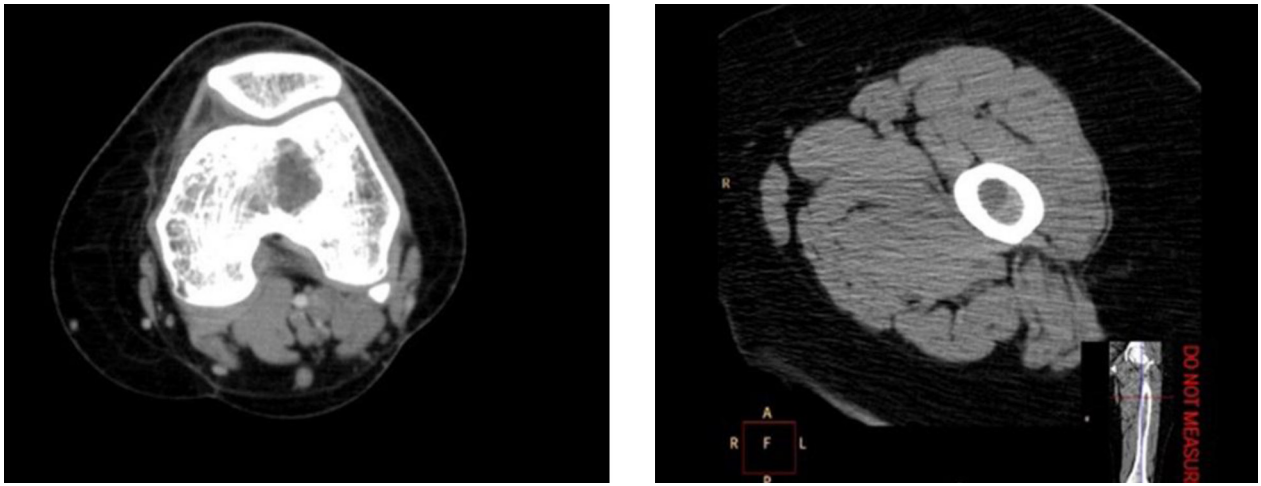
On computerized tomography (CT), a hypo dense 20cm cystic lesion was observed located medullary from the left femur at the intertrochanteric level extending to the distal metaphysis, and the width of the medullar segment with slight lobular contours caused by cortical thinning (Figure 1).

In the magnetic resonance (MRI) examination, an evident hypo intense mass lesion was observed on T1-weighted images, localized in the femur distal metaphyseal-diaphyseal area involving the entire bone medulla and accompanying soft tissue components visualized as evident heterogeneous hyper intensity on the T2-weighted series (Figure 2).

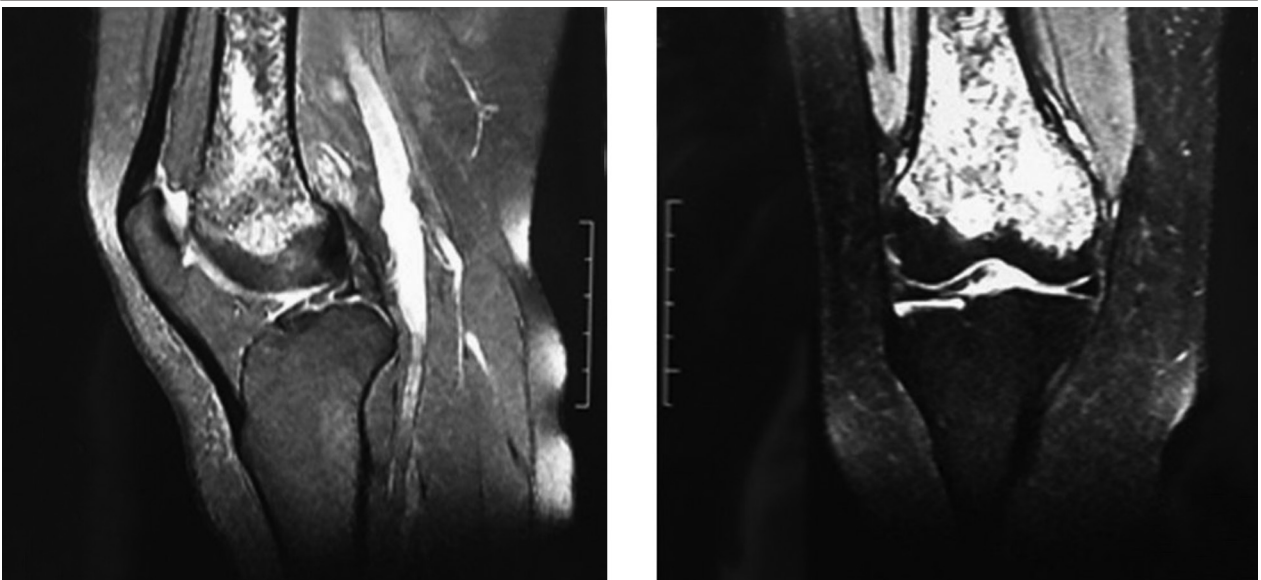
Characteristic of our specific case is a rare involvement of cystic lesion located medullary from the left femur intertrochanteric level extending to the distal metaphysis.

The patient underwent surgery and following intraoperative left femur medullar cyst excision, the histopathological examination determined hyalinized membranous fragments within calcified areas consistent with hydatid cyst (Figure 3). The microbiological examination determined positive titer > 1/280.

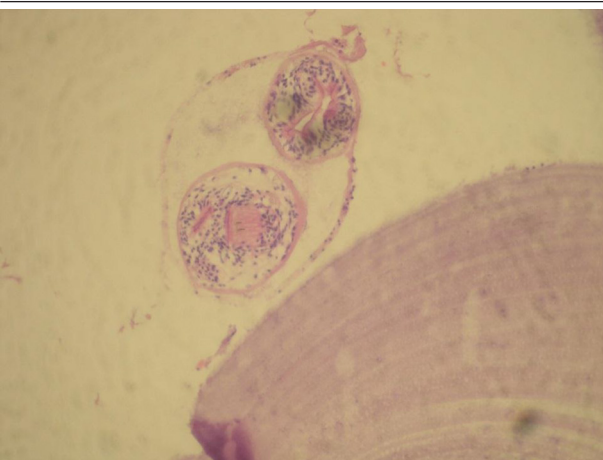
Immobilization was applied for 1 month postoperatively. For a period of 6 months, mebendazole 400 mg orally was administered. No recurrence was determined clinically or radiologically in the 6-month follow-up period. Negative results were obtained from serological examinations (Box 1).



**Figure 1.** A Transverse Slice of the CT Scan Performed Showing a Mass in the Medullary Canal



**Figure 2.** A Coronal and Sagittal Slice of the MRI Showing Multicystic Lesion in the Distal Metaphysis



**Figure 3.** Wall of the Hydatid Cyst Showing the Laminated Membrane With Scolex (H & E,  $\times 400$ )

### 3. Discussion

Echinococcosis, which is known as unilocular hydatid disease or human hydatid disease is caused by the larvae of *E. granulosus*. In the USA, it is seen in cattle and sheep farming areas (Arizona, Utah, New Mexico, and Wyoming). Similar diseases are seen in South America, Europe, and Australia (4). While sheep are temporary hosts, the Canidae family (domestic dogs, foxes, jackals) is definitive hosts. In the presented case, there was a history of a dog bite in 15 years ago.

The liver is most often affected, but other organ systems (kidneys, intestines, central nervous system, and bones) can also be involved. Bone involvement is seen in less than 3% of cases and cystic or lytic lesions occur in trabecular bone, which may extend to other subcortical areas. The ribs, pelvis, and skull are the most frequently involved bones. Long bones may also be affected. Progressive changes and an expanding cystic appearance

**Box 1.** Summary of Clinical Findings of the Studied Case**Clinical Findings**

**Complaints of pain in the distal area of the left thigh, which had been ongoing for approximately 6 months.**

**The patient had a history of a dog bite in the lateral distal left thigh 15 years ago.**

**In the radiographic examination, there was a left femur distal diaphyseal-metaphyseal radiodense area.**

**Cystic lesion was observed located medullary from the left femur intertrochanteric level extending to the distal metaphysis**

**Surgery: histopathological examination determined hyalinized membranous fragments within calcified areas consistent with hydatid cyst.**

**The microbiological examination determined positive titer > 1/1280.**

**No recurrence was determined.**

may resemble tumor formation (5). In our case, location was shown in the left femur, distal intramedullary area, which is a rarely seen location in the long bones. The symptoms of bone hydatid cyst are nonspecific and often missed. This creates difficulties in diagnosis (6, 7). This case was presented at the polyclinic with nonspecific findings in the distal thigh.

Antigen and antibody levels are helpful in diagnosis. By defining the characteristics of the cyst with imaging methods (plain radiographs, CT, MRI and ultrasonography), the treatment can be planned. It is difficult to diagnose bone hydatid cyst disease radiologically because the findings are nonspecific (8). Single or multiple osteolytic lesions and in some cases, cortical thinning, osteosclerosis, and pathological fractures can be seen on radiographs.

As the localization was in the distal femur intramedullary area in the current case, the clinical and tomographic findings indicated a different diagnosis, so initially an intra-osseous ganglion cyst was considered. The diagnosis of hydatid cyst was made with histopathological examination after excision of the mass and the indirect hem agglutination (IHA) test was found positive.

Intra-osseous localization of hydatid cyst disease often involves the spongy area. When the cyst expands, it destroys the surrounding trabecular area and reaches a significant size (3). This condition results in pathological fracture. In the case presented here, the radiological findings were nonspecific.

Therefore, a preoperative diagnosis of skeletal hydatid disease is difficult and accurate diagnosis is generally made during surgery and confirmed histologically (3). In the histopathological examination, the cyst external wall is chitinous and internally the germinal layer is surrounded by a fibrous capsule and granulation tissue (9, 10).

The best treatment for bone infection originating from echinococcosis is removal of the involved bone or amputation. However, curettage and lavage with hypertonic salt, 1% formalin, or 0.5% silver nitrate solutions have been attempted (5). In this case, cyst excision and debridement were applied.

In cases with suspected echinococcosis, diagnostic biopsy or aspiration are contraindications. The surgical approach must be well-planned as drainage of the cyst contents may spread the disease, which must be avoided. If there is leakage, the application of formalin or iodine may reduce the damage of the spread. If a difference is shown in response to treatment, albendazole or mebendazole can be used as medication (5). In this case, albendazole therapy was applied to prevent spread and recurrence. The limitation of our study is the short follow-up period to decide on the prognosis. However, this case of a hydatid cyst located in a long bone was presented to emphasize the need to consider hydatid cyst in the differential diagnosis when a well-defined cystic mass is determined in endemic regions.

### Authors' Contributions

Hasan Onur Arikand and Mehmet Arican: manuscript writing and serving as the surgery team member; Nesibe Kahraman Cetin: pathological evaluation; Umit Sarp: rehabilitation after surgery period.

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