

Case Report

Primary hydatid disease in the femoral condyle: a case report

Yi Zhao^{1*}, Jinlu Yu^{2*}, Jingbo Wang³, Dong Zhu¹, Tiecheng Yu¹

Departments of ¹Orthopedics, ²Neurosurgery, ³Pathology, The First Hospital of Jilin University, Changchun, China.
*Equal contributors.

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Abstract: It is rarely reported that primary femoral hydatid disease results from a dog bite wound contaminated by *E. granulosus* eggs. However, in this case, the primary femoral hydatid disease might occur due to a wound contaminated by *E. granulosus* eggs after the woman was bitten by her neighbor's dog 24 years earlier. The case of a 57-year-old woman suffering from primary osseous hydatid disease in the femoral condyle is presented here. Obtaining the correct diagnosis was difficult. A diagnosis of *Echinococcus granulosus* infestation was made histologically. The femoral osseous hydatid disease was treated by curettage and lavage with hypertonic salt for 10 minutes during surgery. After surgery, albendazole was administered to prevent recurrence (14 mg/kg/d for 6 weeks). The patient was recurrence-free after one year of follow-up. Our experience with this case is helpful. Human *E. granulosus* infection usually occurs after ingestion of food or water contaminated by *E. granulosus* eggs. It is more common from *E. granulosus*-contaminated food than from an *E. granulosus*-contaminated wound. Based on the evidence, we deduced that the primary bone hydatid disease did not result from ingestion of *E. granulosus*-contaminated food or water but instead from an *E. granulosus*-contaminated wound. Therefore, we hope that more clinicians will be able to verify this deduction by clinical observation or by carefully planned animal experiments.

Keywords: Echinococcus, hydatid, femoral condyle, *E. granulosus*

Introduction

Hydatid disease, also called echinococcosis or hydatidosis, is a parasitic disease involving the larval stage of the tapeworm *Echinococcus* [1, 2]. Two types of *Echinococcus* can infect humans: *Echinococcus granulosus* (cystic hydatid disease) and *Echinococcus multilocularis* (alveolar hydatid disease) [1, 3, 4]. The former is prevalent in several areas of the world and is mainly found in sheep-raising areas [3, 4], while the latter is limited to certain areas of central Europe, Canada, and Alaska [4, 5]. In this report, the patient suffered from intramedullary cystic hydatid disease in the left femoral condyle. Osseous cystic hydatid disease is rare, even in endemic areas, contributing to only 0.5-2.5% of all cases [6, 7]. Femoral condyle involvement is extremely rare, and few cases have been published in the literature [8]. In this case, the patient did not live in an endemic area or raise sheep or breed dogs. Her only close contact with a dog occurred 24 years ear-

lier, when she was bitten by her neighbor's dog. This dog bite wound likely led to osseous hydatid cystic disease in her femoral condyle. Here, we presented the likely cause of disease and the clinical, histological, and radiographic findings in a case of primary hydatid disease of the femoral condyle. This information will be helpful for diagnosing and treating this type of patient in the future.

Case report

A 57-year-old Chinese woman was referred to our hospital from a northern rural area of the Jilin province in China. Her neighbor had been diagnosed with brain hydatid disease 4 years earlier and had died 3 years earlier. Two years prior to presentation, the patient experienced an unexpected and severe pain in her right knee while performing routine housework. One year prior to presentation, the right knee pain worsened while walking. Immediately thereafter, she suffered from swelling and functional

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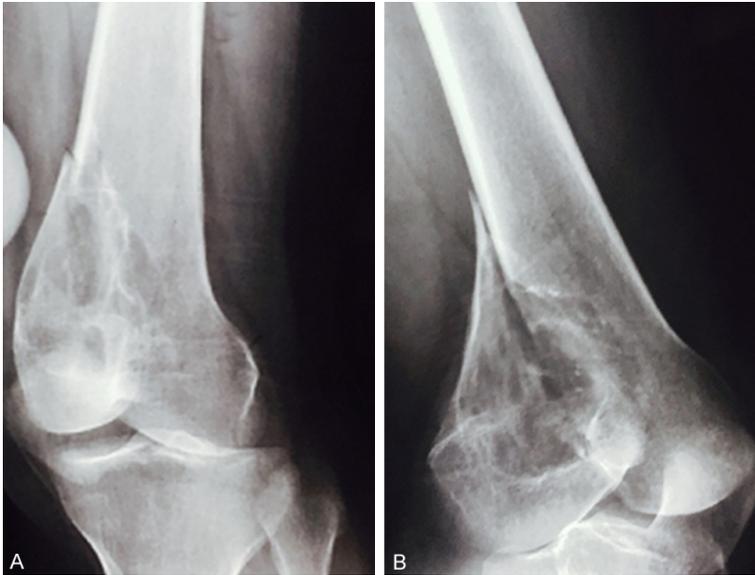


Figure 1. Anteroposterior (A) and lateral (B) x-rays showing a lytic lesion of the medial condyle of the femur.



Figure 2. CT image showing an osteolytic lesion in the left femoral condyle.

impairment of the right knee. She presented at our hospital after 30 hours of travelling. She had no history of fever, malaise, or weight loss. Her medical history was unremarkable. She had a history that included a dog bite to the left upper arm 24 years earlier, resulting in an approximately 5 cm long wound with visible deep muscle. The wound was covered with sugar without cleaning, which was a local rural method used to treat wounds in Northern China. The wound healed in 14 days.

A clinical examination revealed moderate swelling, abnormal movement in the right knee and

serious pain by palpation over the distal part of the femur. She was afebrile and without other signs of infection. The overlying skin was normal. An x-ray examination of the femoral condyle showed lytic lesions primarily affecting the left medial condyle of the femur. The cortex was thinned, and a fracture line that extended from the femoral medial condyle to the medial edge of the distal femur was noted. No sclerosis or periosteal reaction was observed (**Figure 1**). A CT examination of the femoral condyle showed the same result as the x-ray examination (**Figure 2**). However, an MRI examination of the femoral condyle

yielded more detail, showing many similarly sized lytic lesions in the medial condyle of the femur, many of which were filled with abnormal, inordinate, oval-shaped materials, some of which had extended from the fractured area of the femoral condyle to the soft tissue (**Figure 3**). Based on her history and a radiological examination, we assumed that the patient was suffering from a pathological fracture of the left medial condyle of the femur. However, to obtain a precise diagnosis and avoid performing unnecessary surgery, a decision was made to biopsy the left medial condyle of the femur.

The lesion was biopsied under spinal anesthesia. A careful incisional biopsy of the left medial femoral condyle was performed using oncologic principles so as not to preclude subsequent surgical treatment. The dissection was extended to the bones of the femur. After exposition of the fracture site of the cortical bone, several cysts emerged from the medial femoral condyle. The maximum diameter of the cysts was 11 mm, and they contained clear fluid (**Figure 4A**). The cysts and a sample of the left femoral condyle were obtained and sent for pathological evaluation. In a histopathology examination, a lamellar structure of the cystic wall characteristic of *E. granulosus* was observed (**Figure 4B**). The biopsy results confirmed a diagnosis of hydatid cystic disease of the left femoral condyle. A chest x-ray and abdominal

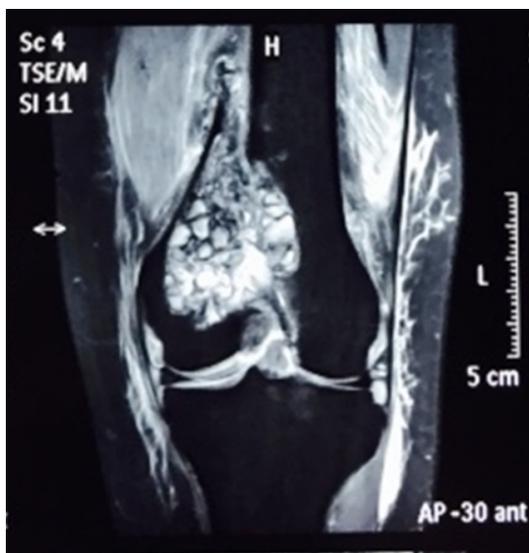


Figure 3. MRI findings reveal a multiloculated hydatid cystic lesion involving the medullary cavity of the left femoral condyle and causing irregular lytic destruction extending from the fracture site into the soft tissues.

ultrasound showed no cystic lesions. The blood profile indicated the absence of eosinophilia. The sedimentation rate was 56 mm/hr. The patient underwent surgery 4 days after the biopsy. A 12-cm longitudinal incision was made on the superior medial aspect of the left knee. The dissection was extended to the bones of the left femoral condyle. After removal of a very thin layer of cortical bone, cysts emerged from a cavity affecting the left femoral condyle. The cysts were evacuated, and bone curettage was performed. The curetted bone consisted of layers of lamellar and necrotic bone, and it was infused with a 10% NaCl solution for 10 minutes. The fractured femoral condyle was reduced to the femur and fixed with cannulated lag screws. The curetted bone cavity of the left femoral condyle was filled with allograft. The fractured femoral condyle was fixed to the left femur with a locking compression anatomical plate, which is typically used to treat distal medial fractures of the left tibia (**Figure 5A, 5B**).

The postoperative course was uneventful. The patient underwent clinical and radiographic follow-up evaluations at 3, 6, and 12 months and yearly thereafter. At 3 months, she was able to return to her usual activities with mild symptoms. At the last follow-up, one year after

the operation, the patient complained only of minor pain after walking for a distance. A radiological examination of the left femoral condyle showed healing of the fracture and no recurrence of the hydatid disease (**Figure 5C, 5D**).

Discussion

Currently, China is considered one of the most important endemic regions of hydatid disease, which is often seen in Western and Northwestern China, the cattle and sheep farming areas [2, 9]. Jilin province is in Northeast China, which is not one of the main endemic areas [9]. Human hydatid disease occurs after ingestion of food or water that contains *E. granulosus* eggs [10]. This occurs principally in areas where dogs are used to maintain herd grazing animals, particularly sheep [10]. Embryos released from the eggs penetrate the intestinal mucosa, enter the portal bloodstream and are carried to the liver, where they are trapped and become hydatid cysts (70%-75% of all cases) [11]. Some larvae reach the lung (15%-25%) and develop into pulmonary hydatids [11, 12].

Skeletal infestation of the cysts occurs by hematogenous seeding. It is generally accepted that human infected with *E. granulosus* by ingestion of food or water contaminated by *E. granulosus* eggs. However, this patient did not raise sheep or breed dogs. Her only close contact with a dog was when she was bitten by her neighbor's dog 24 years earlier. The 5 cm bite wound was not cleaned and was simply treated with sugar. It is possible that the patient was infected by *E. granulosus* via a bite wound contaminated by *E. granulosus* eggs. Additionally, a report exists of a 43-year-old female patient with primary hydatid disease who had a history that included a dog bite 15 years prior to presentation [8]. The embryos released from the eggs penetrate into blood capillaries and enter the normal circulation. Although the chance is small, the embryos in the circulation are more likely to reach the medullary canal of the femoral condyle and spread along cancellous bone than embryos that are released from eggs in the intestinal canal. Because no adventitia is formed in bone, circulating embryos have no way to escape the femoral condyle. The embryos become cysts, which enlarge and give rise to daughter cysts. Due to the rigid structure of cortical bone, the cysts tend to grow slowly and

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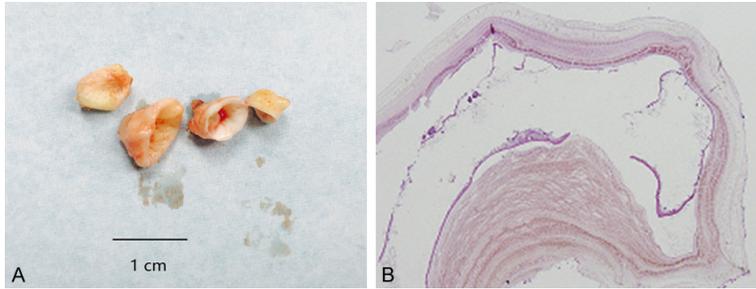


Figure 4. Hydatid cysts (A) obtained from the left femoral condyle during surgery. (B) The lamellar structure of the hydatid cyst wall is shown (hematoxylin and eosin staining, 100x).

seldom exceed 2 cm in diameter. Therefore, unlike lesions in other non-osseous locations, the lesion caused by hydatid cysts in the femoral condyle were polycystic and remained asymptomatic for a protracted period of time (nearly 24 years). The growing cysts caused bone destruction and deformity. Finally, the medial cortex eroded, and the patient suffered from a pathological fracture.



Figure 5. Anteroposterior (A) and lateral (B) x-rays showing the fixation of the femoral fracture 1 day after surgery. Anteroposterior (C) and lateral (D) x-rays of the femoral condyle 1 year after surgery show no recurrence of the hydatid disease in the left femoral condyle.

Radiologically, it is difficult to diagnose hydatid disease in the bone because the radiological findings are nonspecific [13-15]. The radiological images of this patient simply showed lytic lesions in the left medial condyle of the femur where the cortex was thinned. It is difficult to make a differential diagnosis between a giant cell tumor and aneurysmal bone cysts in the femoral condyle because these conditions also express lytic bone lesions with a soft tissue component upon radiological examination. The CT and radiological examinations yielded the same information. However, the findings of the MRI examination were specific, showing many similarly-sized lytic lesions in the medial condyle of the femur. These specific findings differentiate this process from a giant cell tumor or aneurysmal bone cysts in the femoral condyle. Additionally, the MRI examination revealed the extension of abnormal material from the fracture site of the femoral condyle into the soft tissue beyond the femoral condyle. This information indicated that the abnormal material carried a risk of dissemination, sensitization and

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anaphylaxis. Therefore, a diagnostic biopsy was performed. The results of the diagnostic biopsy yielded a final diagnosis of the abnormal material.

The optimal treatment of intraosseous hydatid disease is surgical removal of the involved bone combined with chemotherapy [15-17]. The hydatid cysts infiltrate the cancellous bone without a clear delimitation. Therefore, it is difficult to achieve complete eradication of the hydatid lesion. The local recurrence rate has been reported to be 70%-80% [18, 19]. Ideally, the lesions should be removed using the same technique as for a malignant tumor [16, 19, 20]. Curettage and lavage with hypertonic salt, 1% formalin, or 0.5% silver nitrate solutions have been attempted [20, 21]. After surgery, a chemotherapy (mebendazole or albendazole) regimen that is effective against *E. granulosus* is administered to prevent recurrence [21]. Although no consensus exists regarding the optimal dosage and duration of chemotherapy, albendazole seems to be the most effective drug, particularly when given on a long-term basis [17, 22]. The WHO recommends 10-14 mg/kg/d for 4 weeks of every 6 weeks [10, 12, 23]. Treatment might be continued in some cases for 3 months or longer [12]. Long-term follow-up is necessary before considering bone hydatidosis as being completely healed [24].

Our case, although occurring in a very rare location, is quite typical of osseous involvement. The lesion was asymptomatic for a long time and first became symptomatic when the patient suffered from a pathological fracture. The diagnosis was not made before the diagnostic biopsy. Liver and lung screening were necessary because additional hydatid cysts have been observed in the lung or liver in cases of bone hydatidosis [24]. Such lung and liver cysts might be asymptomatic for several years. In our case, the osseous hydatid disease was treated by curettage and lavage with hypertonic salt for 15 minutes during surgery. After surgery, albendazole was administered to prevent recurrence (14 mg/kg/d for 6 weeks). The patient had no recurrence at the one year follow-up evaluation.

Humans are usually infected by *E. granulosus* following ingestion of food or water contaminated by *E. granulosus* eggs [18, 25, 26]. However, in this case, the primary femoral

hydatid disease might have resulted from a dog bite wound contaminated by *E. granulosus* eggs. Based on the evidence, we deduce that this patient's primary bone hydatid disease might not have resulted from ingestion of food or water contaminated by *E. granulosus* eggs, but rather resulted from a wound contaminated by *E. granulosus* eggs. We hope that more clinicians will verify this deduction by clinical observation or carefully planned animal experiments.

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Disclosure of conflict of interest

None.

Address correspondence to: Tiecheng Yu, Department of Orthopedics, The First Hospital of Jilin University, 71 Xinmin Avenue, Changchun 130021, China. Tel: +86-13596058780; E-mail: tiechengyu@163.com

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