# Primary cystic echinococcosis presented as prolonged disabling knee osteoarthritis: a case report

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## ABSTRACT

Cystic echinococcosis (hydatid disease) is a neglected tropical disease common in Asia, South America and Sub-Saharan Africa. It is a parasitic disease caused by larval stage of *Echinococcus granulosus*. The commonest organs affected by hydatidosis are the liver and the lung. Primary bone hydatidosis in humans is a rare disease accounting for 1–2% of echinococcosis cases. To our knowledge secondary hydatidosis of joints is uncommon and few cases of hydatid disease of the knee joint have been reported.

We present a case of primary cystic echinococcosis of the knee joint occurring in a 43-year-old female presenting mainly as disabling right knee arthritis in the form of chronic (13 years) painful swelling, which ultimately led to her inability to walk. Imaging and histopathologic examination revealed cystic echinococcosis of the right knee joint. Subsequent pulmonary, liver and bone screening for primary lesions were negative. To our knowledge, this is the first ever report on primary knee hydatid disease.

It is possible that pre-existing arthritis modulated the local environment of the knee, rendering it susceptible to hydatidosis. This should be taken into account in the differential diagnosis of arthritis particularly in endemic areas.

**Keywords:** hydatid cyst, knee hydatidosis, echinococcosis, knee osteoarthritis, Sudan

## INTRODUCTION

Echinococcosis is a zoonotic disease caused by infection with the larval stage of a helminth belonging to the genus *Echinococcus.*[1] Although it is rare in Europe and North America, hydatid disease (cystic echinococcosis) is endemic in North Africa and South America.[2] According to the WHO, the incidence rates can exceed 50 per 100000 person-years.[3]

Cystic echinococcosis can occur in a variety of human body sites, most commonly in liver (50–80%) and lungs (15–47%).[4] Few cases of secondary hydatidosis of the joints have been mentioned in literature. This case report describes a rare case of primary distal right femoral hydatidosis with involvement of the right knee joint.

## CASE REPORT

A 43-year-old female from Atbara, northern Sudan, presented with right knee pain which started 13 years earlier. The patient denied any history of trauma to her knee and said she was seen regularly by her physician. The problem was managed as a case of non-specific arthritis. However, the pain increased and became very severe during the last two years. The knee became swollen and the patient developed inability to walk.

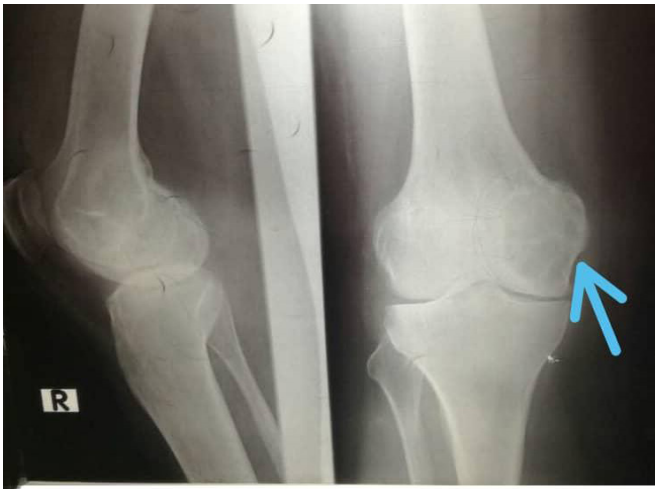


*Figure 1. X-ray of the right knee showing cystic lesion on the right medial condyle*

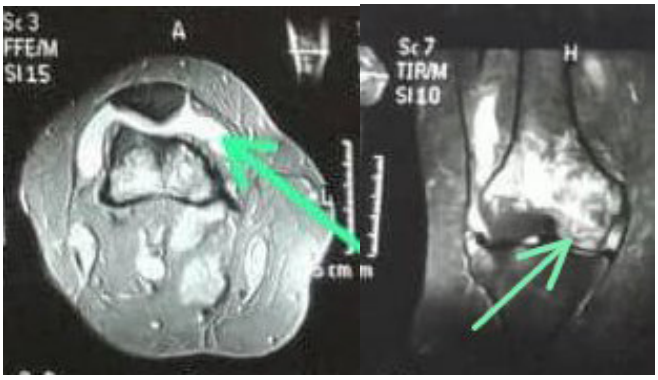


*Figure 2. MRI of (left) the axial plane right knee joint showing joint effusion in T2W and (right) coronal plane of the right knee showing a serpiginous structure of low signal intensity in the medial femoral condyle representing a detached Echinococcus cyst membrane.*

Physical examination of the central nervous system, chest, and abdomen were unremarkable. Local examination showed swelling of the medial aspect of the right knee measuring 8 cm × 10 cm. The swelling was cystic and was not fluctuant or transilluminating. There were no signs of acute inflammation or regional lymphadenopathy on palpation. However, active joint movement was limited to 120°.

Laboratory investigations revealed negative Rheumatoid Factor and normal complete blood count, erythrocyte sedimentation rate, serum uric acid and renal function test. Lateral and anteroposterior X-rays of the right knee were suggestive of a cystic lesion in the right femoral condyle (Figure 1). MRI of the right knee showed a pathologic signal intensity involving the femoral metaphyseal and epiphyseal regions as well as the medial femoral condyle (Figure 2). The lesion was hypo-intense in T1 weighted image and heterogeneously hyper-intense in T2 weighted image with some multifocal cystic appearance. The MRI also showed some trabecular cystic appearance in the posterior aspect of the lateral tibial condyle, intra articular effusion, and marked synovial hypertrophy. These findings further suggested an unusual cause of arthritis in this patient.

The histopathological examination of a core needle biopsy taken from the medial condyle of the right femur showed pink lamellated membranes consistent with an Echinococcus cyst membrane. Based on imaging and histopathology the diagnosis of hydatid disease of the right knee was reached.

One month later the patient underwent right knee exploration under spinal anaesthesia via a medial parapatellar approach. Intraoperatively, synovial thickening was noted. Extended curettage was done and bone cement was applied to compensate the bone defect. Curettage products were sent for histopathologic examination and the patient was started empirical antibiotics and analgesics. Microscopic examination of curettage products showed necrotic bony tissue with lamellated pinkish membranes and scolices along with extensive neutrophilic infiltrate and occasional calcification. This confirmed the initial diagnosis of hydatid disease of the distal femur and the knee joint. (Figure 3).

Abdominal and lung CT scan done on the patient revealed no abnormality and confirmed the notion of a primary disease of distal femur with knee joint involvement.

The patient was prescribed albendazole tablets 400mg daily for three months. On follow up after six weeks, the patient showed marked improvement. Her knee pain, swelling and tenderness all resolved. She started to regain her ability to walk. Radiological imaging showed complete resection and filling of the defect with cement and no signs of recurrence (Figure 4). Unfortunately, one month later she developed an acute drug induced hepatitis. Albendazole was withdrawn in anticipation of her recovery in order to put her on a second line antihelminthic agent. Regrettably, the patient failed to return for follow-up.

## DISCUSSION

Hydatid disease of bone and joints represent less than 0.5% of human echinococcosis.[5,6] The most frequently involved osseous areas are the vertebral column, the pelvis, the long bones, and the skull. Joint involvement usually occurs secondary to the involvement of the nearby osseous tissue.[7]

Several cases of secondary hydatidosis with joint involvement have been reported in the literature. These include knee hydatidosis extending from femur bone from primary liver lesions;[8] hip joint due to extension from the sacrum;[9] knee hydatid disease with tibial and femoral lesions secondary to hepatic hydatidosis;[10] lumbosacral

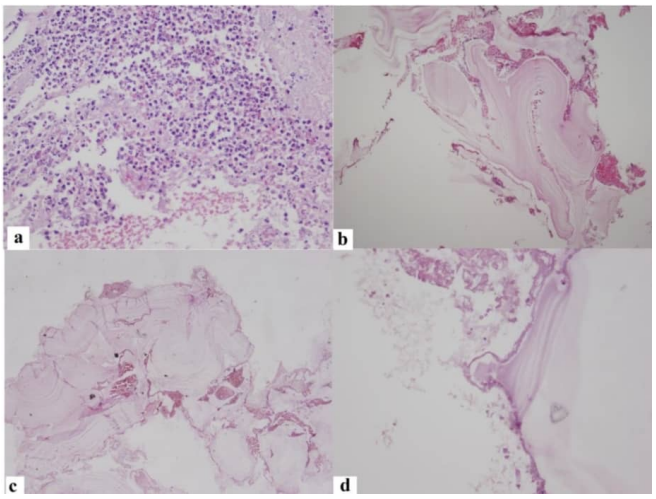


*Figure 3. a) Necrosis and neutrophilic infiltration, haematoxylin and eosin stain (H&E) x4. (b) and (c) Lamellated membranes, H&E x40. (d) Lamellated membranes and scolices, H&E x60.*

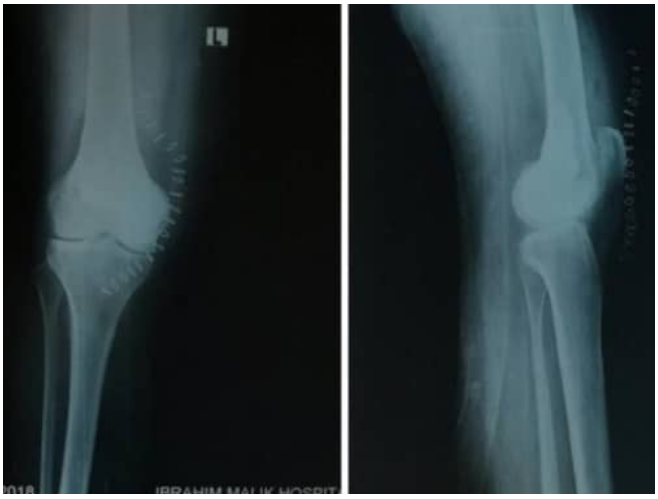

*Figure 4. Follow up X-ray after surgery showing cement covering the gap after surgical removal of the cystic*

joint involvement secondary to lung and liver hydatid disease; hip hydatidosis with nearby osseous destruction; shoulder joint secondary to liver hydatid disease;[5] and knee hydatidosis secondary to primary bone hydatidosis of distal femur.[11]

In contrast to the secondary hydatid disease cases reported in the literature, our case is unusual and possibly unique in that there was no primary lesion detected in the liver, lungs or brain. The only abnormality in this case was in the distal part of the right femur. Another unusual feature of our case is that the patient presented with a chronic history and total loss of ability to move the right knee joint because of pain and later swelling. This could be attributed to the slow growth and progression of the lesion. The unusual presentation mimicking osteoarthritis of the right knee perhaps initially inhibited the correct diagnosis. On histopathological examination the patient was noted to have extensive neutrophilic infiltration of the hydatid cyst membranes that involved the adjacent bony tissue. This could be attributed to post-biopsy secondary bacterial infection that resolved following removal of the infected tissue and administration of cephalosporin antibiotics.

An important lesson from this case report is that biopsies in cystic lesions are important and can help in reaching the correct diagnosis. A possible explanation of the primary knee hydatidosis is that the patient was initially suffering from another type of arthritis which damaged her knee joint and compromised blood supply to the area.[12]

The patient is originally from an area well-known for breeding livestock including camels, the herbivore most infected with hydatidosis in Sudan.[13] Some people in Sudan have the practice of eating raw camel liver. When exposed to the organism, possibly through ingestion of raw camel meat, the larvae settled in her distal femur and subsequently caused knee hydatidosis.

Known side effects of albendazole use include gastrointestinal disturbances, thirst, dizziness, headache, alopecia and itching. However, it may rarely induce hepatitis.[14] This unlucky patient developed increased liver enzymes forcing treating physicians to stop albendazole and give her follow-up appointment. Unfortunately, the patient did not return for the appointment.

## CONCLUSION

Hydatid disease can present as chronic arthritis. Diagnosis depends on radiological and histopathological examination. Misdiagnosis or delayed treatment may lead to significant disability. Hydatid disease of a joint should be included in the differential diagnosis of arthritis with adjacent bone cysts in endemic areas.

**Conflicts of interest:** none

**Acknowledgment**

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