

Case Report

Bone hydatid disease of distal femur and diaphysis of tibia: report of two cases with review of literature

Ramesh Kumar*, Chandrashekhar M. Badole, Girish Mote, Gajanan D. Chintawar

Department of Orthopaedics, MGIMS, Sewagram, Wardha, Maharashtra, India

Received: 26 December 2016

Accepted: 02 February 2017

*Correspondence:

Dr. Ramesh Kumar,

E-mail: rkhatanamgims@mgims.com

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ABSTRACT

Cystic echinococcus or hydatidosis is a parasitic infection of humans and animals. In this we are reporting 2 cases. one is hydatid disease at distal femur left side and other patient having hydatid disease at shaft tibia rt side. both cases investigated and definitive surgery was done as arthrodesis with long tibio-femoral nail for distal femoral hydatidosis and curettage for tibial hydatidosis. Both patient given albendazole and followed up. in recent follow-up both cases do not have signs of recurrence. hence case report of these two cases is discussed along with review of literature.

Keywords: Arthrodesis, Curettage, Hydatid disease, Long bones

INTRODUCTION

Cystic echinococcus or hydatidosis is a parasitic infection of humans and animals. This parasite has 12 different types although 4 types cause disease in humans: *Echinococcus granulosus*, *Echinococcus multilocularis*, *Echinococcus vogeli*, and *Echinococcus oligarthrus*.¹

Hydatid cyst occurs in many areas of the world but it is especially endemic in South America, Asia, Australia, North and East Africa, and Middle East where sheep and cattle breeding is a common occupation.² Incidence of hydatid cyst disease ranges between 3 and 50 per 100,000 population.^{3,4}

Although hydatid cyst can form disease in any organ except hair and nails, it usually affects liver, spleen, and lungs.⁵ Involvement of these 3 organs is responsible from 90% of infections.⁶

Primarily bone involvement of cyst hydatid occurs in 1% to 2.4% and among them 30% to 50% involves the vertebrae, 15% involves pelvis, and less frequently involves long bones.^{3,7}

CASE REPORT

Case 1

50 year old female presented to Orthopedics OPD with complains of pain and swelling over left side knee and thigh since 5 year. On physical examination, there was pain on deep palpation and diffuse swelling over the distal area of the left thigh and knee as per Figure 1.

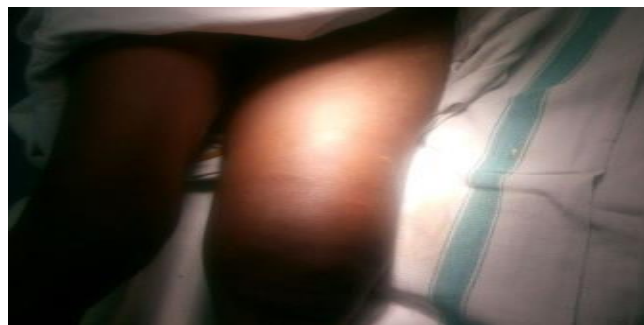


Figure 1: Diffuse swelling over the distal area of the left thigh and knee.

No pathological lymph node was determined on palpation of surface lymph nodes.

In the laboratory tests, no pathology was determined in routine haemogram and bio-chemical tests. In the radiographic examination, there was lytic area in distal femur left side as per Figure 2.



Figure 2: Lytic area in distal femur left side.

In the magnetic resonance (MRI) examination, Large altered signal intensity lesion involving distal diaphysis, metaphysis of shaft of femur involving bilateral femoral condyles, showing heterogeneously enhancing solid core with chondroid type of matrix mineralization and multiple peripheral cysts as visible in Figure 3.

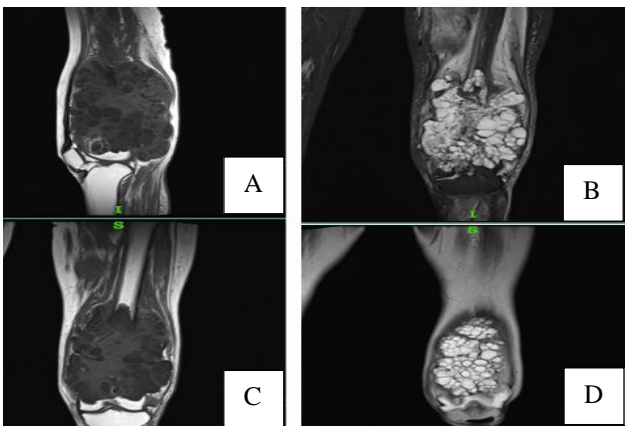


Figure 3: Lesion involving distal diaphysis, metaphysis of shaft of femur.

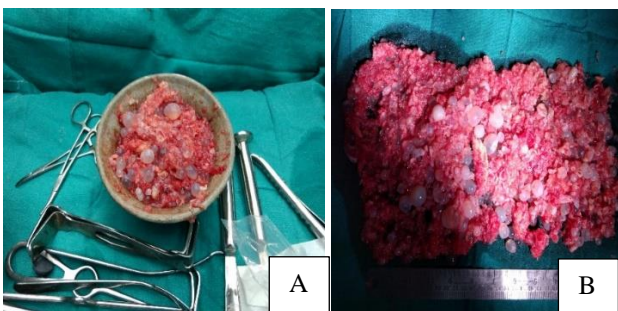


Figure 4: Intraoperative cyst excision material.

The patient underwent diagnostic incisional biopsy surgery and following intraoperative left femur cyst excision (Figure 4), the histopathological examination determined hyalinized membranous fragments within calcified areas consistent with hydatid cyst as per Figure 5.

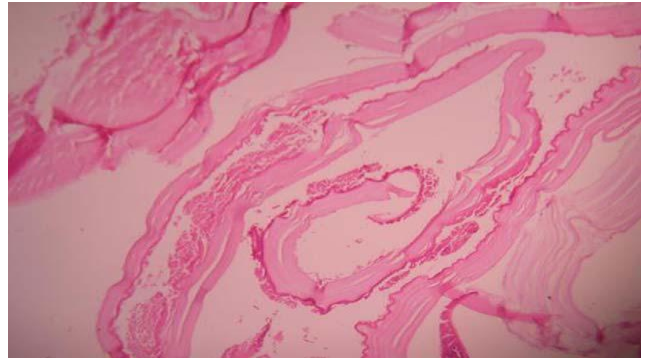


Figure 5: Histopathological diagram.

Immobilization was applied for 1 month by upper tibial skeletal traction postoperatively. Albendazole 400 mg orally was administered.^{8,9} No recurrence was determined clinically.

Then after 1 month, definitive surgery done as Arthrodesis of knee joint fixed with tibio-femoral interlocking nail (Figure 6) with both cortical and cancellous bone grafting harvested from bilateral fibula and bilateral iliac crest.¹⁰ On day 3 post op, Patient was mobilized on crutches non weight bearing. Patient discharge dafter removal of stitches and in recent follow-up there was no recurrence.

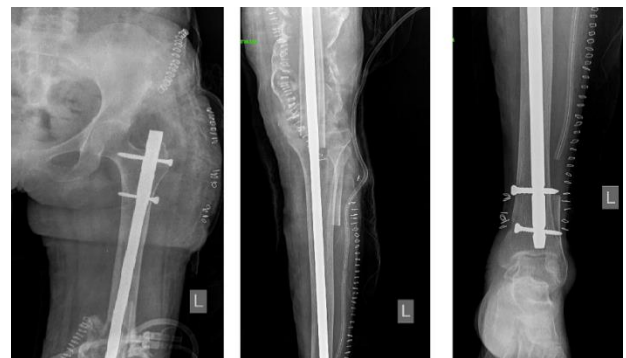


Figure 6: Arthrodesis of knee joint fixed with tibio-femoral interlocking nail with cortical and cancellous bone grafting.

Case 2

17 year old male presented with pain and swelling over right leg since 2 months. On physical examination, there was tenderness with diffuse swelling over right leg. No pathological lymph node was determined on palpation of surface lymph nodes.

In the laboratory tests, no pathology was determined in routine haemogram and bio-chemical tests. Then radiograph was taken which showed osteolytic lesion at shaft of Right tibia (Figure 7).



Figure 7: Osteolytic lesion at shaft of right tibia.

MRI was done which reported as metaphyseal fibrous defect of right tibia (Figure 8). Thereafter biopsy was taken and histopathology report confirmed as hydatid cyst disease. After this definitive surgery was done and all cysts were removed and curettage done (Figure 9).

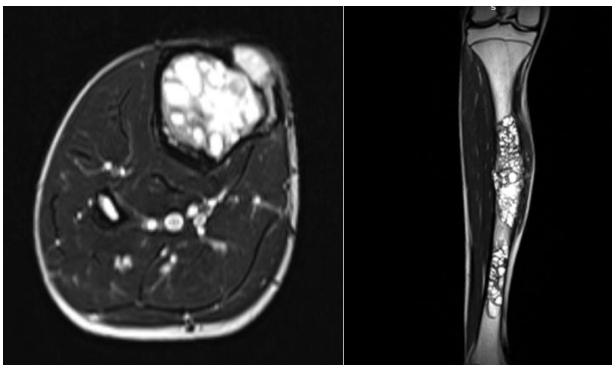


Figure 8: MRI picture shaft of right tibia.

Patient was started on Albendazole for 3 months and long leg slab applied to Right lower limb. Patient was kept non-weight bearing for 2 months. Then patient was followed up for 6 month (Figure 10) and in recent follow-up there was no recurrence.



Figure 9: Immediate post-op radiograph.

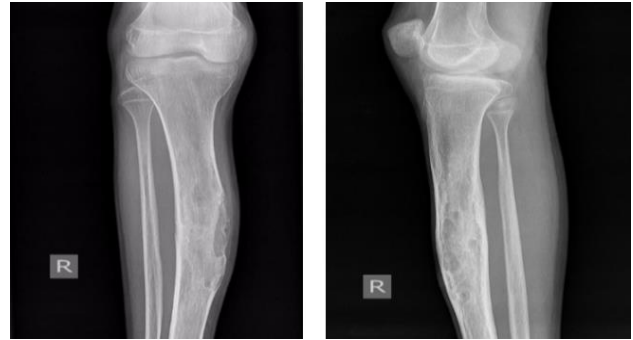


Figure 10: Follow-up Radiograph at 6 months.

DISCUSSION

Hydatid disease is caused by the tapeworm *Echinococcus*. Species of the genus *Echinococcus* include *Echinococcus vogeli*, *Echinococcus granulosus* and *Echinococcus multilocularis*. In man and domestic animals, this parasitic infection is most commonly caused by the larval stage of *Echinococcus granulosus*.^{11,12} The adult worm resides in the intestine of the canine, which functions as a definitive host. Ingestion of ova passed with the feces from the definitive host by man and domestic animals as intermediate hosts, hatch in the small intestine and enter the blood circulation, locating in different tissues where they produce hydatid cysts.³ These cysts commonly occur in the lungs and liver, but can be found in any other organ or tissue including bones, spleen, heart, eye, brain, and genitourinary tract. Anatomoclinical changes are peculiar to localization in the bone.¹¹ From the anatomopathologic stand point, this localization marks the torpid, insidious progression of the parasite into the bone tissue, leading to a diffuse, extensive, invasive process; so from the clinical stand point, wherever it is localized, its complete surgical eradication is rarely possible.¹¹

Hydatid cysts of bone remain asymptomatic over a long period, and are usually detected after a pathologic fracture or secondary infection, or following the onset of compressive myelopathy in cases of vertebral lesions.¹³ However, a definite preoperative diagnosis without histological examination is often difficult as there are no pathognomonic signs, radiological findings may be confused with those of other tumoral lesions, and serological tests are of limited value.¹¹ When a pathological fracture occurs in long bones due to hydatid cyst, non-union is common. The present case might have been harboring asymptomatic infestation for quite a long time and it was only pain that brought the patient to us for treatment. *Echinococcus* joint disease is usually due to secondary extension from an adjacent bone. Transarticular extension from the pelvic bone to the femur or sacrum, similar to the present case, has been reported in the literature.^{17,18}

The threat of anaphylactic reaction to the cyst fluid and spread of infection to the surrounding structure and soft

tissues, sometimes by the seeding of hooklets, adds anxiety to both open biopsy and definitive surgery. Difficulty in both diagnosis and management are hallmarks of hydatidosis of bone. The disease mimics chronic osteomyelitis, fibrous dysplasia of bone osteosarcoma, benign cystic lesion of bone, brown tumor (hyperparathyroidism), and various other neoplastic lesions.¹⁹⁻²¹ It should be considered in the differential diagnosis of expansile osteolytic lesions, especially in endemic areas.^{21,22} A conclusive diagnosis of hydatidosis has only been reached in half of the cases preoperatively.¹³ The indirect hemagglutination test is more reliable in diagnosing the condition than the Casoni skin test or Weinberg's complement fixation test.^{17,23} Histopathological examination is confirmatory.²¹ The anaphylactic nature of the cyst fluid demands strict precautions and skillful handling during biopsy and definitive surgery.

Osseous hydatidosis should be treated with radical resection with a wide margin of healthy tissue. This may be difficult, but incomplete removal results in recurrence.¹⁵ Radical resection in the pelvis and hip is extremely challenging, and total eradication of parasitic osteitis is almost impossible.^{13,17,24} joint reconstructive surgeries after radical excision are almost technically impossible in the pelvis and hip, although in the past, hip arthroplasty and custom-made prosthesis have been tried.^{3,4,7,16} Extensive surgical approaches are always accompanied by the dangers of recurrence and infections. Even patients in a good general condition may not tolerate such surgeries. Sepsis may be a cause of death.^{16,24}

Overall, a review of the literature reveals a poor prognosis if the disease is extensive in the pelvis and proximal femur.^{19,20} for distal femur more follow up studies needed. The purpose of this article is to alert orthopedic surgeons of this morbid condition and to emphasize the fact that this disease should be suspected in cystic lesions affecting any organ of the body in painful swellings, pathological fractures with non-union, especially in endemic areas of the world. Early diagnosis helps in eradication and salvage of the bone; misdiagnosis and delayed diagnosis are always fraught with the danger of amputation, recurrence, and sepsis.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: Kumar R, Badole CM, Mote G, Chintawar GD. Bone hydatid disease of distal femur and diaphysis of tibia: report of two cases with review of literature. *Int J Res Med Sci* 2017;5:1126-30.