Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/apme

Case Report

Hydatid disease of bones – Imaging spectrum

A. Pudhiavan [a,*], Alka Agrawal [b], Prem S. Tripathi [c]

[a] Registrar, Department of Radiodiagnosis, Apollo Heart Centre, Chennai 600006, India
[b] Associate Professor, Department of Radiodiagnosis, MGMMC & MY Hospital, Indore 452001, India
[c] Assistant Professor, Department of Radiodiagnosis, MGMMC & MY Hospital, Indore 452001, India

ARTICLE INFO

Article history:
Received 30 October 2012
Accepted 9 November 2013
Available online 23 November 2013

Keywords:
Hydatid cyst
Bone hydatid
Long bone hydatid
Sacral hydatid

ABSTRACT

Hydatid cyst, caused by echinococcus granulosa, can produce tissue cyst everywhere in body. Skeletal cystic lesion is rare specially in long bones like femur and because of its unusual presentation, its diagnosis may easily be missed, unless be kept in mind. Hydatid disease is a parasitic infestation of the humans that can practically affect any part of the body. It commonly affects liver, lung and muscles. Bone hydatid is less common occurring in only 0.5–2% of cases and humans act as an intermediate host. In the patient evaluated by us, the plain x-ray showed multiple osteolytic lesions along the lower one third of the femur and the upper end of tibia. There was also cortical destruction of the joint surfaces that is seen with involvement of the joint. USG revealed a well-defined anechoic cystic lesion in the region of the calf in the substance of gastrocnemius muscle with fine septation and fine echogenic debris in the dependent part. MRI further provided data on the involvement of bone marrow and the extent of the disease. In the second patient, there was hydatid affection of the sacrum and spinal canal. The diagnosis was confirmed on tru-cut biopsy from the affected bone. Primary hydatid disease is a very rare entity and can be seen without evidence of any other visceral involvement. Cautious suspicion of the disease and early diagnosis is important for the management.

Copyright © 2013, Indraprastha Medical Corporation Ltd. All rights reserved.

1. Introduction

Human echinococcosis is a zoonotic infection caused by the tapeworm of the genus *Echinococcus*. Species of medical importance in humans are *Echinococcus granulosus*, causing cystic echinococcosis (CE); *Echinococcus multilocularis*, causing alveolar echinococcosis (AE); and *Echinococcus vogeli*. *E. granulosus* is the most common of the three. The adult worms inhabit the small intestine of the dog and the ova are shed in its excreta. The ova, when swallowed by sheep, cattle or man, enter the portal venous system and are carried to the various organs of the body, particularly the liver and lungs, where they form cysts. The cysts contain in their walls masses of worm heads or scolices, which if swallowed by the dog develop into adult worms, thus completing the life cycle. Hydatid disease affects the bones in 0.5–2% of cases of which spine is involved in approximately 45% of cases.[1–3] In the vertebral column it affects the lumbar, thoracic and cervical region, in decreasing order of frequency.[3] Affection of long bones like in our case is pretty rare which can involve tibia, femur and practically any long bone.

* *Corresponding author.*
E-mail address: drpudhiavan@gmail.com (A. Pudhiavan).
0976-0016/$ – see front matter Copyright © 2013, Indraprastha Medical Corporation Ltd. All rights reserved.
http://dx.doi.org/10.1016/j.apme.2013.11.003

Fig. 1 – A 50-year-old patient with hydatidosis of femur and tibia. There are multiple cystic lesions with bone destruction seen in the lower end of femur and tibia with destruction of the cortex and involvement of the knee joint cavity. A co-existent hydatid cyst of the calf muscle is also seen with daughter cysts. (a) Coronal T1W MRI, (b) Coronal STIR MRI, (c) Coronal CT reformation, (d) Axial STIR MRI, (e) Axial T2W MRI, (f) Axial CT scan.

1.1. *Case: 1*

A 50-year-old female presented with swelling of the lower one third of the right thigh and the calf region for the past three years, which has been slow growing. The patient was subjected to plain radiography, USG of both abdomen and thigh and MRI of the thigh region. A logiq 3 USG Doppler machine and a 1.5 T SIEMENS Magnetom Avanto machine were used.

In our case, X-ray examination revealed osteolytic lesion of the lower femur also involving the upper tibia crossing the knee joint. Plain CT sections show well-defined lytic lesions with sclerotic margins and erosions involving the cortex. Prominent cortical breach was also noted in our case. The bone disease was also associated with a pathological fracture of femur with extensive bone destruction. T1WI showed multiple intramedullary hypo intense lesions and T2W images showed intramedullary and intramuscular lesions with characteristic hydatid sand. There was associated destruction of the articular surface of knee joint was also noted (Fig. 1).

1.2. *Case: 2*

A 30-year-old female presented with deformity of the lower back. MRI examination of the spine and pelvis was performed. MRI revealed multiple well-defined expansile T2 hyperintense lesions seen in the lumbosacral region. The lesions were causing bony destruction. There were also lesions seen inside the spinal canal. These lesions showed multiple thin walled septations and numerous daughter cysts. No evidence of any marrow edema or any inflammatory reaction was noted. Histopathological examination of the lesions revealed the typical layers of a hydatid cyst with numerous daughter cysts. There were also numerous intramuscular lesions that were seen (Fig. 2).

2. Discussion

Hydatid disease of long bone is a very rare condition. Thomas JD collected twenty-eight examples, mostly from isolated reports in the literature. The most detailed account of hydatid disease of bone is that of Ivanissevich who published a review

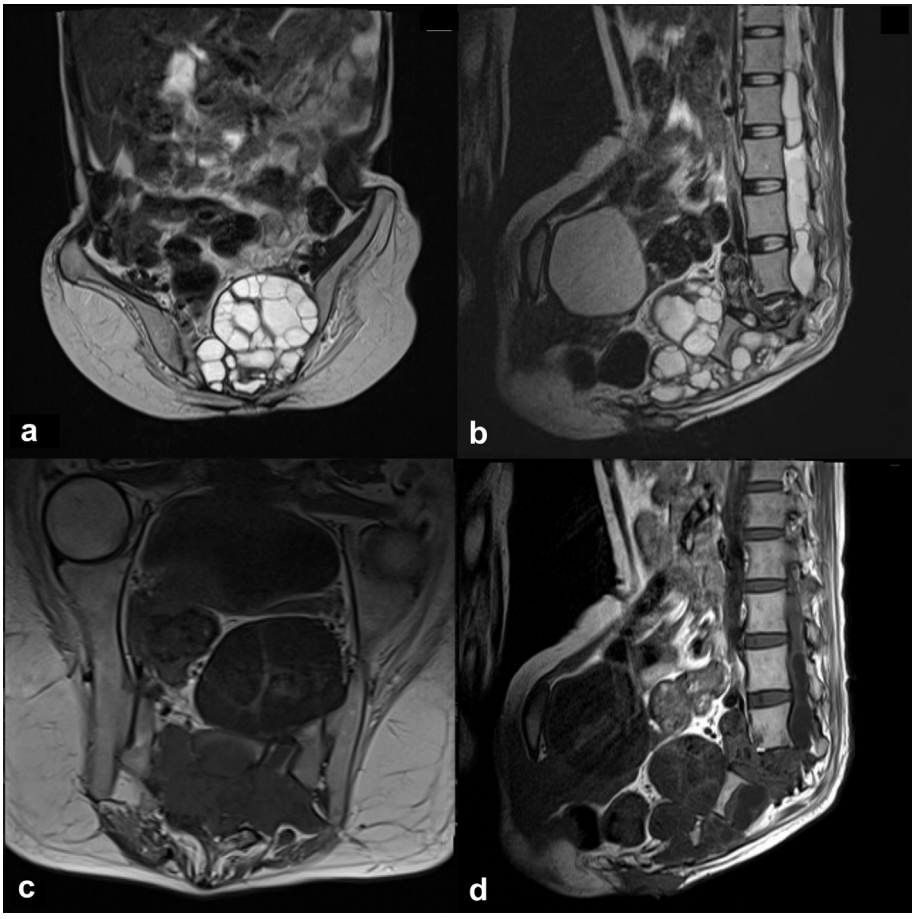

Fig. 2 – A 30-year-old patient with hydatid disease of the lumbo-sacral spine. The MRI images show multiple well-defined cystic lesions in the sacrum and in the lumbar spinal canal with almost complete destruction of the sacrum leading to a spinal deformity. The lesions also cause compression of the cauda equine nerve fibers. (a) Coronal T2W MRI, (b) Sagittal T2W MRI, (c) Axial T1W MRI, (d) Sagittal T1W MRI.

of the subject and described forty-seven cases.[3] In humans, hydatid disease involves the liver in approximately 75% of cases, the lung in 15%, and other anatomic locations in 10%. The frequency of osseous involvement in hydatid disease is 0.5%–2%. It is most commonly seen in the spine and pelvis, followed by the femur, tibia, humerus, skull, and ribs.

Osseous foci may be manifested as pain and deformity, particularly in 30–60 years old age group. Hydatid disease of bone is rarely seen in childhood. The hydatid cyst lie dormant in the bone for as long as 40 years and most skeletal hydatid cyst cases have been adults. Skeletal cystic echinococcosis lesion may be single or multiple.[4,5] As hydatid disease of bone remains asymptomatic over a long-period, it is usually detected after a pathological fracture or secondary infection or following the onset of compressive myelopathy in cases of vertebral lesions.

The initial location of the lesion in long bones is metaphyseal or epiphyseal, later extending to the diaphysis. In bone involvement, pericyst formation does not occur, thereby allowing aggressive proliferation in an irregular branching fashion along the line of least resistance, especially the bone canals.[3,6,7] The parasite replaces the osseous tissue between trabeculae due to the slow growth of multiple vesicles. With time, the parasite reaches and destroys the cortex, with subsequent spread of the disease to surrounding tissues. Extraosseous cysts may calcify, whereas intraosseous disease rarely demonstrates calcification.[2,5] The findings in our case were very similar.

The differential diagnosis of skeletal cystic echinococcosis includes other infectious lesions like tuberculosis, fibrous dysplasia, tumors like simple bone cyst, aneurismal bone cyst, plasmacytoma, osteosarcoma, chondrosarcoma, chondromyxoid fibroma, lymphoma, giant cell tumors, brown tumor and metastases. Simple bone cysts are sharply demarcated lucent lesions with no periosteal reaction whereas aneurismal bone cysts are expansile osteolytic lesions, with thin sclerotic margins and demonstrate the characteristic fluid–fluid levels. Giant cell tumor and brown tumors are difficult to differentiate from hydatid disease and may need serological confirmation. Osteosarcoma and chondrosarcoma are bone forming aggressive tumors with wide zone of transition where as hydatid causes bone lysis by the parasite.

The most common radiological manifestation of skeletal hydatid disease is a lucent expansile lesion with cortical thinning. Surgery is the treatment of choice for hydatid bone lesions.[8] Many authors have advocated wide resection of the involved bone along with the surrounding soft tissue as the only definitive treatment of the condition with or without

chemotherapy using albendazole or mebendazole.[8] In conclusion, the preoperative differential diagnosis of skeletal cystic lesions should include cystic echinococcosis, since the diagnosis may easily be missed, unless be kept in mind.

Conflicts of interest

All authors have none to declare.

REFERENCES

1. Merkle E, Schulte M, Vogel T. Musculoskeletal involvement in cystic echinococcosis. Report of eight cases and review of literature. *AJR Am J Roentgenol*. 1997;168:1531.
2. Torricelli P, Martinelli C, Biagini R, Ruggieri P, De Cristofaro R. Radiographic and computed tomographic findings in hydatid disease of bone. *Skeletal Radiol*. 1990;19:435–439.
3. Engin G, Acunas B, Rozanes I, Acunas G. Hydatid disease with unusual localization. *Eur Radiol*. 2000;10:1904–1912.
4. Sapkas SG, Stathakopoulos DP, Bibis GC, Tsarouchas JK. Hydatid disease of bones and joints. *Acta Orthop Scand*. 1998;69:89–94.
5. Morris B, Madiwale C, Gary A, Chavhan G. Case report: hydatid disease of bone. *Australas Radiol*. 2002 Dec;46(4):431.
6. Fanian H, Marnani KM. A case report of hydatid disease in long bone. *J Res Med Sci*. 2005;10(2):01–04.
7. Arti H, Darani HY. Fibular hydatid cyst. *Indian J Orthop*. 2007;41(3):244–245.
8. Zlitni M, Ezzaouia K, Lebib H, Karray M, Kooli M, Mestiri M. Hydatid cyst of the bone: diagnosis and treatment. *World J Surg*. 2001;25:75–82.