

Pelvic bone and hip joint hydatid disease revealing a retroperitoneal location

Abdelhalim El Ibrahim, ¹ Amal Ankouz, ² Abdelkrim Daoudi, ¹ Abdelmayid Elmrini ¹

¹Department of Orthopaedic Surgery B4, University Hospital of Fez, Morocco;

²Department of Digestive Surgery C3, University Hospital of Fez, Morocco

Abstract

Echinococcosis is a parasitic disease produced by the larval stage of *Echinococcus granulosus*. Hydatid disease of bone is rarely seen in humans and it has been reported in only 1-2% of cases of echinococcosis. We present a patient who developed hydatid disease of the left pelvic and femoral bones with cartilage destruction of the ipsilateral hip joint revealing a retroperitoneal location of hydatid cyst. Hydatid bone must be present in the differential diagnosis of chronic monoarthritis. Skeletal involvement is usually secondary to visceral hydatidosis that we must research. Early diagnosis allows eradication and salvage of the bone and the hip joint. Delayed diagnosis is always fraught with the risk of recurrence and sepsis.

Introduction

Hydatid cyst disease is a parasitic disease caused by a cestode known as echinococci. The genus *Echinococcus* includes three species; *Echinococcus multilocularis*, *Echinococcus vogeli* and *Echinococcus granulosus*. *Echinococcus granulosus* is the most common cause of hydatid disease in man.^{1,3} The definitive hosts are dogs, foxes, and other carnivores. The tapeworms live in the small bowel of these hosts and infected ova are shed in the feces. When ingested by intermediate hosts such as man, sheep, or cattle, the larvae enter the portal circulation. The larvae eventually reach the liver; where most of them are trapped. Sometimes, larvae reach the lungs and other areas of the body and form cysts. The life cycle is completed when the definitive hosts consume infested viscera of the intermediate host.

Bone lesions are reported in only 1-2% of cases.⁴ The strong structure of osseous tissue limits the growth of the hydatid cyst, which spreads along medullar and trabecular channels. The trabeculae are slowly resorbed due to pressure without any cortical extension.

The cysts extend to surrounding soft tissues if the bone cortex is eroded. The disease affects long bones, vertebral column, pelvis, and costae in order from least to most affected region.^{1,5} (Table 1). We report a case of hydatid disease of the left pelvic and femoral bones with cartilage destruction of the ipsilateral hip joint revealing a retroperitoneal location of hydatid cyst.

Case Report

A 35-year old woman presented with one year history of pain in her left hip. There was no history of prior trauma. On physical examination, there was pain and limitation on motion of the left hip. Treatment that included anti-inflammatory drugs and exercise did not alleviate the pain.

Laboratory studies revealed normal results. A simple radiograph of hip showed intraosseous cystic lesions in the left pelvic bone and femur with uniform space narrowing of the left hip (Figure 1).

Our patient was admitted initially for a probable coxarthrosis. Because the radiological aspect was suspect, a femoral biopsy was performed confirming the presence of *Echinococcus granulosus* within the osseous and synovitis lesions. Magnetic resonance imaging (MRI) showed multiple cystic lesions were present in the left pelvic bone, femoral bone and pelvic cavity (Figure 2).

The retroperitoneal hydatid cyst was managed by aspiration. The patient received mebendazole 40 mg/kg/day which was well tolerated. She referred to only mild pain in her left thigh. A new simple radiograph after three months showed multiple cystic lesions involving the left pelvic bone and enlargement of lesions (Figure 3). She has received albendazole 10 mg/kg/day with no changes in clinical symptoms and mild enlargement of the cysts on imaging evaluation. A Girdlestone arthroplasty was suggested but our patient doesn't believe in this option.

Discussion

Echinococcosis is a parasitic disease produced by the larval stage of echinococcus granulosus. Hydatid disease of bone is rarely seen in humans and it has been reported in only 1-2% of cases of echinococcosis.⁴ The vertebral column, the pelvis, the long bones and the skull are most commonly involved.^{7,8}

Hydatid cysts of bone remain asymptomatic over a long period, and are usually detected after complication or at an advanced stage when lesions have become extensive.⁹ There

Correspondence: El Ibrahim Abdelhalim, Department of Orthopaedic Surgery B4, UH Hassan II of Fez, Fez, 30000, Morocco
E-mail: halimibrahimi@yahoo.fr

Key words: hydatid bone, joint involvement, retroperitoneal hydatid cyst, surgery.

Received for publication: 7 April 2009.

Revision received: 20 May 2009.

Accepted for publication: 26 May 2009.

This work is licensed under a Creative Commons Attribution 3.0 License (by-nc 3.0)

©Copyright A. El Ibrahim et al., 2009
Licensee PAGEPress, Italy
Orthopedic Reviews 2009; 1:e8
doi:10.4081/or.2009.e8

are a variety of complications when osseous involvement is present; they include deformity, pathological fractures, secondary infection or neurological problems, such as paraparesis.

Hydatid disease joint involvement is usually due to secondary extension from the adjacent bone, although primary hydatid synovitis after hematogenous spread of the infection can be seen.¹⁰

Diagnosis of the hydatid bone disease is primarily based on findings of X-ray and CT scans.

The radiological signs include lucent osseous lesions associated with expansion of the bone and thinning of the cortex. In patients with these signs, soft tissue calcification is highly suggestive of hydatid disease. Although hip joint destruction is uncommon, it can be seen, as in our case when pelvic and femoral bones are involved. MRI and computed tomography are valuable in delineating the extent of bone and soft tissue abnormalities.¹¹ The diagnosis is often made on the basis of the characteristic radiographic appearance of the lesions.

However, a definite pre-operative 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

Hydatid bone must be present in the differential diagnosis of chronic monoarthritis.¹² Infestation of hip prosthesis has been reported.¹³ Giant cell tumors, solitary bone cysts, aneurismal bone cysts, fibrous dysplasia, bone metastasis, neurofibromatosis, chronic osteomyelitis, tuberculosis of the bone, brown tumor (hyperparathyroidism), and various other neoplastic lesions should be considered in the differential diagnosis of osseous hydatid disease.⁹

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.^{14,15} We present a patient who developed hydatid disease of the left pelvic and femoral bones with cartilage destruction of the ipsilateral hip joint revealing a retroperitoneal location of hydatid cyst.

In our case, hydatid bone disease was suspected from radiologic findings. Femoral and synovitis biopsy confirmed the presence of *Echinococcus granulosus* within the osseous and synovial lesions.

The literature offers various treatment approaches.^{2,16-18} Until recently, the basic treatment has been surgical excision or resection (Table 2). Unfortunately, the results have been discouraging.^{9,23} Complete surgical excision with a wide margin of healthy tissue and curettage is the basic approach for osseous hydatid disease. This surgery may be difficult, with a high risk of recurrence. Radical resection in the pelvis and hip is extremely challenging and total eradication of parasitic osteitis is almost impossible.^{17,23}

Reconstructive surgeries after radical excision are almost technically impossible in the pelvis and hip, although in the past, hip arthroplasty² and custom-made prosthesis⁸ have been tried. 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.^{14,24} Overall, a review of the literature reveals a poor prognosis if the disease is extensive in

Table 1. Bone manifestation of Echinococcus.⁶

Location	Percentage
Spine	30%
Pelvis/hip	20%
Femur/tibia	15%
Humerus	15%
Ribs/scapula	10%
Phalanges	5%
Head	5%

Table 2. Cases of pelvic and hip hydatid disease, treatment methods and outcomes.

Authors	Bone localization	Number of cases	Treatment	Complication	Outcome
Siwach 2009 ¹⁹	Femur and pelvis	1	Amputation and chemotherapy	Sepsis and death	-
Khan 2008 ²⁰	Pubis, femur and pelvis	1	Chemotherapy	-	Unsatisfactory
Masse 2004 ¹⁸	Ischion, hip and pelvis	1	Surgery	-	Satisfactory
Martinez 2001 ²¹	Pelvis and hip	8	Surgery and chemotherapy	Chronic lesions 37%	Satisfactory 63%
Bulzunegui 1997 ¹⁰	Pelvis, femur and hip	1	Surgery and chemotherapy	-	Satisfactory
Wirbel 1995 ⁹	Pelvis, femur and hip	1	Total hip replacement and chemotherapy	Recurrence and instability of hip	Unsatisfactory
Agarwal 1992 ²²	Pelvis, femur and hip	2	Surgery and chemotherapy	-	Satisfactory

the pelvis and femur.^{19,22,24,25}

Isolated medical therapy is not adequate for controlling the process, but it can be added to

surgery or, used like isolated therapy when complete excision is not possible.^{2,26} Mebendazole, albendazole, and antihelminthic



Figure 1. A simple radiograph of hip showed intraosseous cystic lesions in the left pelvic bone and femur with uniform space narrowing of the left hip.

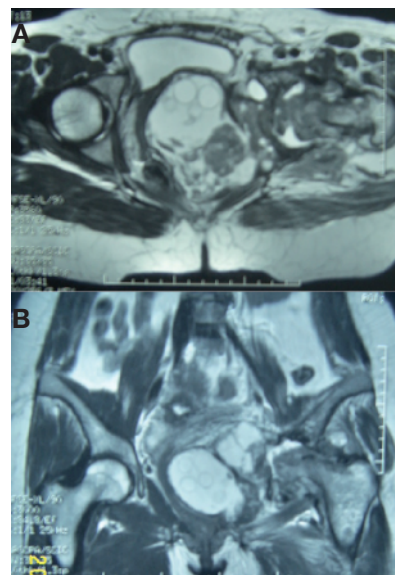


Figure 2. Magnetic resonance imaging (MRI) was performed, multiple cystic lesions were present in the left pelvic bone, femoral head and pelvic cavity. (A). Horizontal image. (B). Coronal image.

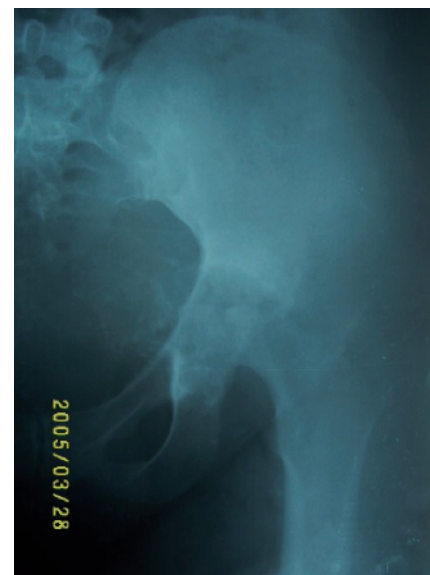


Figure 3. Simple radiograph three months later, enlargement of the lesions with pelvic deformity.

drugs are used for chemotherapy. Albendazole has been found to be better absorbed than mebendazole and exhibits superior efficacy against helminths.² The aim of this work is to alert orthopedic surgeons to this morbid condition and to emphasize the fact that this disease should be suspected in cystic osseous lesion of hip, especially in endemic areas of the world. Skeletal involvement is usually secondary to visceral hydatidosis that we must research. Early diagnosis allows eradication and salvage of the bone and the hip joint. Delayed diagnosis and misdiagnosis are always charged with the risk of handicap, recurrence, and sepsis.

References

1. Agarwal S, Shah A, Khadi SK, Rooney RJ. Hydatid bone disease of the pelvis: a report of two cases and review of the literature. *Clin Orthop* 1992; 280:251-5.
2. Szypryt EP, Morris DL, Mulholland RC. Combined chemotherapy and surgery for hydatid bone disease. *J Bone Joint Surg* 1987; 69B:141-4.
3. Uzel M, Hiz M, Aydingoz O. Curettage, local heat and cement application in echinococcosis of bone. Presented at the Joint Meeting of EMSOS-AMSTS, Florence, Italy 1995.
4. Argenson C, Griffet J, Lacour C, et al. Vertebral hydatid cyst. A propos of 2 cases. *Rev Chir Orthop Rep Appar Mot* 1989;75:267-70.
5. Porat S, Joseph KN. Hydatid disease of bone: a case report and review of the literature. *Isr J Med Sci* 1978;14:223-7.
6. Schneppenheim M, Jerosch J. Echinococcosis granulosa/cystica of the tibia. *Arch Orthop Trauma Surg* 2003;123:107-11.
7. Chikhaoni N, Adil A, Iladiri R. Radiological appearance of vertebromedullary hydatidosis. Report of 12 cases. *J Radiol* 1993;74: 621-8.
8. Jlidi R, Yaakoubi MT, Ladeb MF, et al. L'hydatidose osseuse. *Ann Pathol* 1992;12: 98-101.
9. Wirbel RJ, Mues PE, Mutschler WE, Salomon-Looijen M. Hydatid disease of pelvis and femur. A case report. *Acta Orthop Scand* 1995;66:440-2.
10. Belzunegui J, Maiz O, Lopez L, Plazaola I, Gonzalez C, Figueroa M. Hydatid disease of bone with adjacent joint involvement: a radiological follow up of 12 years. *Br J Rheumatol* 1997;36:133-5.
11. Torricelli P, Martinelli C, Biagini R, et al. Radiographic and computed tomographic findings in hydatid disease of bone. *Skel Radiol* 1990; 19:435-9
12. Campoy E, Rodreguez Moreno J, Del Blanco J, et al. Hydatid disease: An unusual cause of chronic monoarthritis. *Arthritis Rheum* 1995;38:1338-9.
13. Voutsinas S, Sayakos J, Smyrnis P. Echinococcus infestation complicating total hip replacement: a case report. *J Bone Joint Surg* 1987;69A:1456-8
14. Sapkas GS, Stathakopoulos DP, Babis GC, Tsarouchas JK. Hydatid cyst of the bones and joints: 8 cases followed for 4-16 years. *Acta Orthop Scand* 1998;69:89-94.
15. Migotti B, Tomaseo I, Poljanic S, Marjanović D. Importance of scintigraphy in the treatment of hepatic echinococcus. *Acta Chir Iugosl* 1977;24:151-3.
16. Ozkan H, Dogramaci Y, Kose O, et al. Primary hydatid cyst of the humerus. *Ann Acad Med* 2008;37:440-1.
17. Morris B, Madiwale C, Garg A, Chavahan GB. Case report: hydatid cyst of bone. *Aust Radiol* 2002;46:431-4.
18. Massè A, Parola PG, Brach del Prever EM, Gallinaro P. Hydatidosis of the pelvis: a case report with a 25-year follow-up. *Arch Orthop Trauma Surg*. 2004;124:203-5.
19. Siwach R, Singh R, Kadian VK, et al. Extensive hydatidosis of the femur and pelvis with pathological fracture: a case report. *Int J Infect Dis* 2009 Mar 31. [Epub ahead of print]
20. Khan JA, Devkota P, Acharya BM, et al. Bony hydatid disease of superior pubic ramus with extension into pelvis and proximal thigh. *J Nepal Med Assoc* 2008;47: 139-41.
21. Martinez AA, Herrera A, Cuenca J, Herrero L. Hydatidosis of the pelvis and hip. *Int Orthop* 2001;25:302-4.
22. Agarwal S, Shah A, Kadhi SK, Rooney RJ. Hydatid bone disease of the pelvis. A report of two cases and review of the literature. *Clin Orthop* 1992;280:251-5.
23. Meziante A, Bachechar N, Benkirane A, Ouadfel J. Hydatid cyst of pelvis: a case report. *Acta Orthop Belg* 1987;53:517-9.
24. Herrera A, Martinez AA. Extraspinal bone hydatidosis. *J Bone Joint Surg Am* 2003; 85:1790-4.
25. Merkle EM, Schulte M, Vogel T, et al. Musculoskeletal involvement in cystic echinococcosis: report of eight cases and review of the literature. *Am J Roentgenol* 1997;168:1531-4.
26. Winning A, Braslins P, McCarthy JS. Case report: nitazoxanide for treatment of refractory bony hydatid disease. *Am J Trop Med Hyg* 2009;80:176-8.