Case Report

Bone echinococcosis with hip localization: A case report with evaluation of imaging features

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Abstract

Hydatid disease (HD) is a zoonotic parasitic disease caused by the larvae of Echinococcus. Bone echinococcosis is rare, accounting for 0.5% to 4% of all echinococcosis. We describe a particular case of pelvic echinococcosis. A 29-year-old man initially presents with pain in his left hip for several years. After an accidental fall from a tree, he suffered a fracture of the left acetabulum. X-rays and CT scans showed an osteolytic area of the acetabulum with bony cortical interruption. MR imaging demonstrated extensive area of osteostructural alteration of the iliac wing and the left acetabulum due to multiple cysts with enhancement of the walls after administration of Gadolinium-based contrast agents. A CT-guided biopsy of an osteolytic area was performed with diagnosis of echinococcus cyst. He underwent albendazole therapy and subsequently echinococcus cyst exeresis, bone curettage, and left hip arthroplasty. Twenty-two months after surgery, CT scan showed recurrence of disease. After 4 years and 6 months of chronic therapy CT scan showed an increase in size of the cyst at the site of the disease recurrence. Five years and 4 months after the first operation, a new cyst exeresis and pelvic bone curettage with implant retention was performed. This case report demonstrates that hydatid cysts should be considered as a possible cause for non-specific pelvic pain, especially in endemic locations.

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Introduction

Human echinococcosis is a zoonotic infection. Cystic echinococcosis may affect all organs, but mostly settles in the liver and lungs. Primary Echinococcus infection of bone is rare, with reported rates of 0.5%-4% of the total cases of hydatid disease [1]. The spine is the most common osseous location of infection, followed by the long bones (femur, tibia, and humerus), skull, and ribs. Pelvic involvement is extremely

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Case report

A 29-year-old man initially presents with pain in his left hip for several years. After an accidental fall from a tree, he suffered a fracture of the left acetabulum treated with traction for 30 days. He underwent X-rays and CT scans that showed an osteolytic area of the acetabulum with bony cortical interruption in a pathological acetabular fracture. A CT-guided biopsy of an osteolytic area was performed with diagnosis of echinococcus cyst.

He underwent albendazole therapy for 5 months and subsequently echinococcus cyst exeresis, bone curettage, and left hip arthroplasty with cemented Burch-Schneider ring via Smith-Petersen approach were performed, continuing albendazole for 4 weeks after surgery.

Twenty-two months after surgery, CT scan with contrast medium showed the appearance of fluid-like formation starting from osteolysis of the posterior column of the acetabulum suggesting recurrence of disease, therefore, albendazole therapy was resumed with subsequent reduction of the cystic area.

After 4 years and 6 months of chronic therapy an additional control CT scan was performed showing an increase in size of the cyst at the site of the disease recurrence.

Five years and 4 months after the first operation, a new cyst exeresis and pelvic bone curettage with implant retention via xipho-pubic approach was performed, considering the exclu-

Fig. 1 - X-rays demonstrates an expansile lytic process involving the acetabular region of the left hip.

Fig. 2 - CT scan demonstrates a lytic process involving the acetabular region of the left hip and also shows extravascular fluid/overfluid component with multiple contextual calcifications involving the gluteus minimus muscle and the ipsilateral internal obturator muscle.
sively endopelvic extension of the recurrence with involvement of the obturator fossa.

From a radiological point of view, radiographs demonstrated an expansile lytic process involving the acetabular region of the left hip (Fig. 1).

CT scan confirmed the lytic process that also shows extrasosseous fluid/overfluid component with multiple contextual calcifications involving the gluteus minimus muscle and the ipsilateral internal obturator muscle; the intramedullary margins of the process were not well defined (Figs. 2 and 3).

MR imaging demonstrated extensive area of osteostructural alteration of the iliac wing and the left acetabulum due to multiple cysts with enhancement of the walls after administration of Gadolinium-based contrast agents (Fig. 4), apparently communicating with the joint. Within the larger cysts, a few millimetric oval images with a void signal in the joint cavity are appreciated. This formation extended medially into the pelvis – with slight dislocation of the iliac vessels – and laterally to the gluteal region, contained below the fascial planes where the largest cysts (max. 68 × 50 × 76 mm) are appreciated. Associated medullary edema of the femoral head and, to a lesser extent, of the left iliac wing adjacent to the sacroiliac synchondrosis was found, correlating with reactive-phlogistic phenomena. STIR-sequences were also performed that demonstrated hyperintensity of the iliac muscle and the gluteus medius muscle at their bony insertion (Fig. 5). Radiographs performed after surgery showed the results of curettage and prosthetic implantation (Fig. 6). A CT scan performed 22 months after first surgery showed the recurrence of the disease starting from the posterior acetabular wall and involving soft tissues medial to the coxofemoral joint (Fig. 7). After the second surgery no residual disease was detectable on CT scan (Fig. 8).

**Discussion**

Bone hydatidosis is uncommon and, in contrast to other organs, the hydatid cysts in bone do not develop a pericyst, which is a dense fibrous protective layer of modified host cells [2]. The rigid structure of bone limits uniform enlargement and leads to the slow and insidious, yet extensive, spread of
Fig. 5 – MRI with STIR sequences demonstrates multiple cystic areas at the site of the left acetabular region, the largest of which was 68 × 50 × 76 mm, with some parietal calcifications. There is associated medullary edema of the femoral head and, to a lesser extent, on the left iliac wing adjacent to the sacroiliac synchondrosis, in relation to reactive-phlogistic phenomena. In addition hyperintensity of the iliac muscle and the gluteus medius muscle at their bony insertion.

Fig. 6 – X-rays after cyst exeresis with implantation of left total hip prosthesis.
Fig. 7 – CT axial and coronal reconstruction demonstrates recurrence of echinococcus disease starting from the posterior acetabular wall and involving soft tissues medial to the coxo-femoral joint.

Fig. 8 – CT scan post-excision of the disease recurrence with implant retention.

diagnosis of bone echinococcosis. Definitive diagnosis can be made with biopsy or aspiration, but the latter has classically not been recommended because of the risk of further dissemination or anaphylaxis. Antibody analysis, which was done in this patient, may also be helpful.

The primary treatment option, regardless of the location of the infection, is usually surgery. Other treatment options include aspiration with instillation of alcohol, physiologic saline, or antiparasitic medication. Systemic pharmacotherapy is also usually administered. The most commonly administered drug is albendazole, with praziquantel being an alternative choice. Pharmacotherapy is started before surgery and can be continued afterward, in some cases indefinitely [2,4].

Recurrence is frequent, as it happened in this case, and it reaches rates of 30%-100% [14]. This is particularly true in bones. As mentioned above, unlike other viscera, there is no adventitia formation in skeletal hydatidosis (due to the rigid structure of the bone). This allows for microvesicular invasion of the daughter cysts into the bony tissue [12]. As a result, due to this issue and lack of clear-cut demarcation between normal and pathological tissues, complete eradication of the parasite is often unsuccessful despite extensive surgery. Presentation within an anatomically complex district such as the pelvis can only further decrease the chances of obtaining radical surgery. Unfortunately, due to its high rate of recurrence prognosis is poor. Long-term radiologic follow-up is recommended at least once a year to identify recurrence at the site that needs to be excised.

This report has tried to emphasise how this disease requires awareness and up-to-date knowledge on the part of physicians, especially in endemic areas when patients present with generalized musculoskeletal disorders.

**Patient consent statement**

Patient was informed of the publication of a case report regarding his clinical case and provided written consent for pub-
liciation. Furthermore, the patient is aware that his name will not appear in the text of the article.

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