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Case report

Multiple pelvic and thigh hydatid cyst: a case report

Maryem IKEN[1], Adil LAMKHANTAR[2], Mohamed BOUSSAIDANE[2], Hafida NAOUI[1], Badr LMIMOUNI[1], Mostapha BOUSSOUGA[2]

[1]Parasitology and Mycology Laboratory, Mohamed V Military Teaching Hospital, Mohamed V University, Rabat, Morocco
[2]Department of Orthopaedic Surgery and Traumatology, Mohamed V Military Teaching Hospital, Mohamed V University, Rabat, Morocco

Corresponding Author: Maryem Iken; e-mail: bousiken@yahoo.fr

ABSTRACT. Bone cystic echinococcosis is rare disease and its management remain difficult to treat due to frequent recurrences, related to certain locations such as proximal femur and ilium, where radical surgery is difficult to achieve. Medical therapy using Mebendazol can be an ultimate way to insure stabilization and remission of the parasitosis. We present a case of a 37-year-old male who had complaints of pain in left hip and limp four years after surgical management of pertrochanteric fracture of his right lower limb. The first clinical and radiological diagnosis was osteoarthritis of proximal femur and hip. However, higher imaging modalities revealed the diagnosis of hydatid disease of hip with extensive pelvic hydatid cyst localizations. The patient underwent anthelminthic chemotherapy. At 7 years follow-up, the lesions remains stable and the patient has tolerate hip pain, support and walking are impossible without crutches. Hydatid disease of the proximal femur and pelvis must be kept in mind in the differential diagnosis of pathologies of hip-like septic arthritis or tuberculosis. Prolonged medical therapy with anthelmintic medications can constitute an alternative to surgery at very high risk and for extensive lesions where surgery can not achieve carcinologic goals.

Keywords: hydatid disease, pelvis, femur, *Echinococcus*

Introduction

Bone localization of the hydatid cyst (*Echinococcus*) is a rare form of the disorder, often associated with other clinical manifestations. We report a femur upper end case with massive extension to the pelvis, diagnosed late after four years of a pertrochanteric fracture osteosynthesis, and whose the pathological aspect of the bone had gone annoticed on radiological analysis. Isolated medical treatment was chosen given the local extension of the affection and allowed lesional stabilization with a follow-up of seven years.

Case presentation

We report the case of a 37-year-old patient who was admitted to the trauma emergency room for a pertrochanteric fracture managed by Dynamic Hip Screw device (DHS) (Fig. 1a), the pathological aspect of the bone had gone annoticed on radiological analysis. The evolution was marked three months later by development of a sepsis with a fistula bringing back serous fluid from the operative scar, associated with screw migration of the DHS device (Fig. 1b). Operative samples with ablation of the material and abundant washing were carried out (Fig. 1c), the bacteriological and histological study revealed an infectious process with *Staphylococcus* sensitive to Methycillin. The evolution was marked by the drying up of the infection under a double antibiotic treatment based on ciprofloxacin and clavulanic acid, walking was possible with lameness and tolerable pain.

Four years later, the patient consulted following the worsening of the pain in the hip and the appearance of a skin fistula on the external face of the latter. Bacteriological samples made it possible to isolate the same germ (sensitive *Staphylococcus aureus*). Standard X-rays of the hip showed

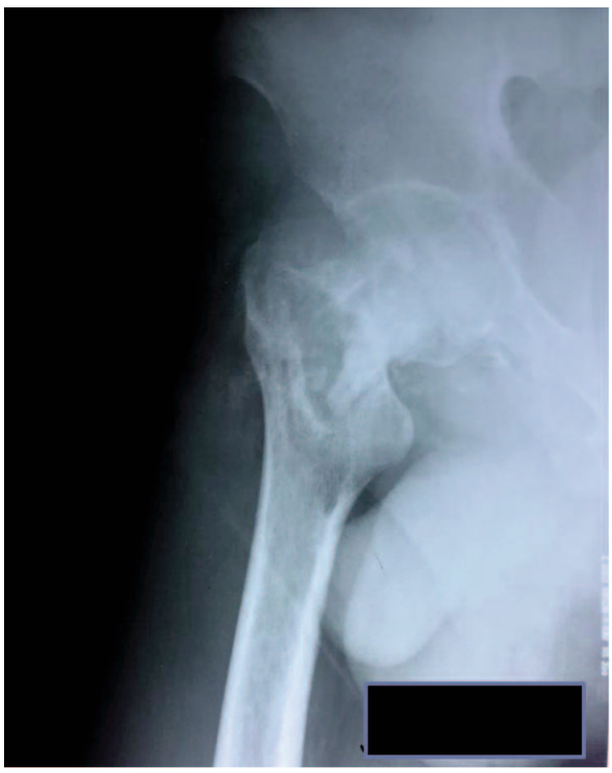

Figure 1a. Misdiagnosed pathological proximal femur fracture

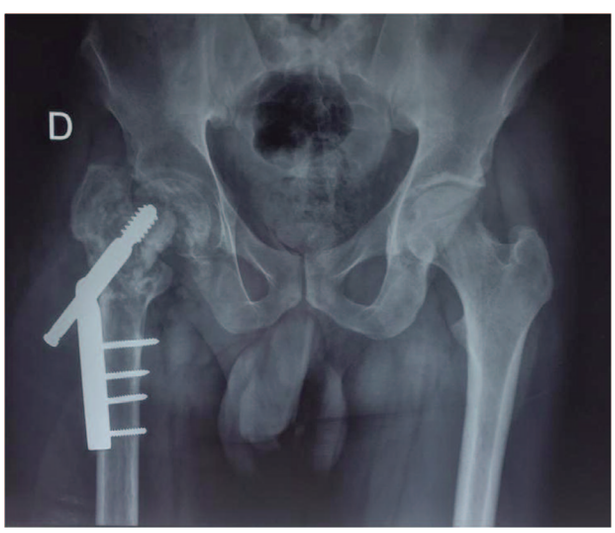

Figure 1b. Screw displacement of the DHS device

osteonecrosis of the femoral head with radiological signs of septic osteoarthritis of the hip, and lytic images extended to the upper 1/3 of the femoral shaft.

Nuclear magnetic resonance imaging has shown the existence of a multivesicular lytic process, centered on the right hip, taking the metaphysis and the proximal 1/3 of the femoral shaft with a pathological fracture. This pathological process extended to the soft parts of the thigh, the

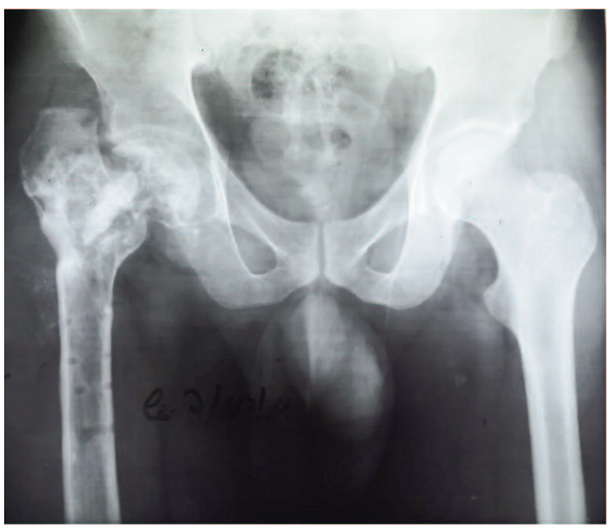

Figure 1c. Radiological aspect after DHS removal

acetabulum and the pelvis, with the presence of multiple vesicular images exerting a mass effect on the rectum, bladder and iliac vessels (Fig. 2a,b). The diagnoses of a septic osteoarthritis with multiple intrapelvic abscesses of bacterial origin were raised, and given the endemic geographical situation of the country, a tuberculous or hydatid origin were also suspected.

Tuberculous serologies were negative, and the biological diagnosis of hydatid involvement was strongly suspected on the result of the serological examinations (Western Bloot and ELISA). Hypereosinophilia was noted on the blood count.

Surgical procedure was then proposed to the patient with a partial hemipelvectomy and an extensive resection of the femoral shaft, associated with the removal of the pelvic vesicular formations. Secondary restoration procedure by massive prosthetic surgery could be performed later depending on the local course of the disease. The surgical risks were explained to the patient who had preferred the alternative of medical treatment with Mebendazole for suppressive purposes, The treatment was administered in courses of 28 days with a break of 14 days between each course, at a dose of 800 mg per day, or 1 tablet of 400 mg twice a day.

After 7 years of follow-up, the patient still had hip pain, he was functionally handicapped with walking in monopodal support, the MRI check shows lesional stability. (Fig. 3a,b).

Discussion

Bone hydatidosis, a rare form of the disease (1 to

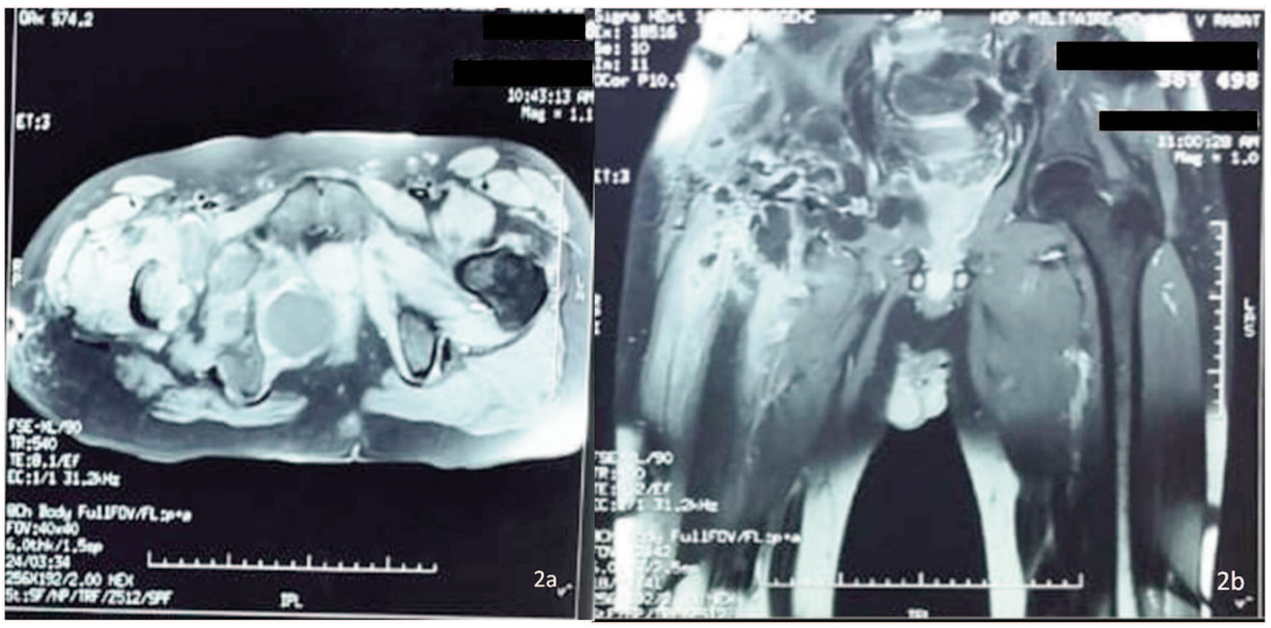

Figure 2. Multiple hydatid cysts on the proximal aspect of femur and pelvic involvement

4%) [1,2], has serious mechanical consequences for the patient due to the major functional handicap it causes [3,4], and vital risk linked to a dangerous localization near vasculo-nervous and visceral structures. Diagnosis is often late, even in endemic breeding areas, given the very slow progression of bone damage [5–7] which can manifest itself in chronic pain or result in a pathological fracture whose management is difficult [8]. This pathology must be raised in the same way as osteoarticular tuberculosis, clinical signs are not specific and the biology not always contributory and can however show a hypereosinophilia [9] and a positive serology in the tests by Western Bloot and ELISA.

Computed tomography (CT scan) and nuclear magnetic resonance imaging (MRI) make it easy to suspect the diagnosis [10] and establish the surgical indication, therapeutic choice as well as the approach. The vertebral, pelvic and upper extremity locations of the femur seem preferential for parasitosis, the radiological aspect is that of significant bone destruction with the presence of well-enveloped cystic images of varying sizes, scattered around the neighboring muscles and soft

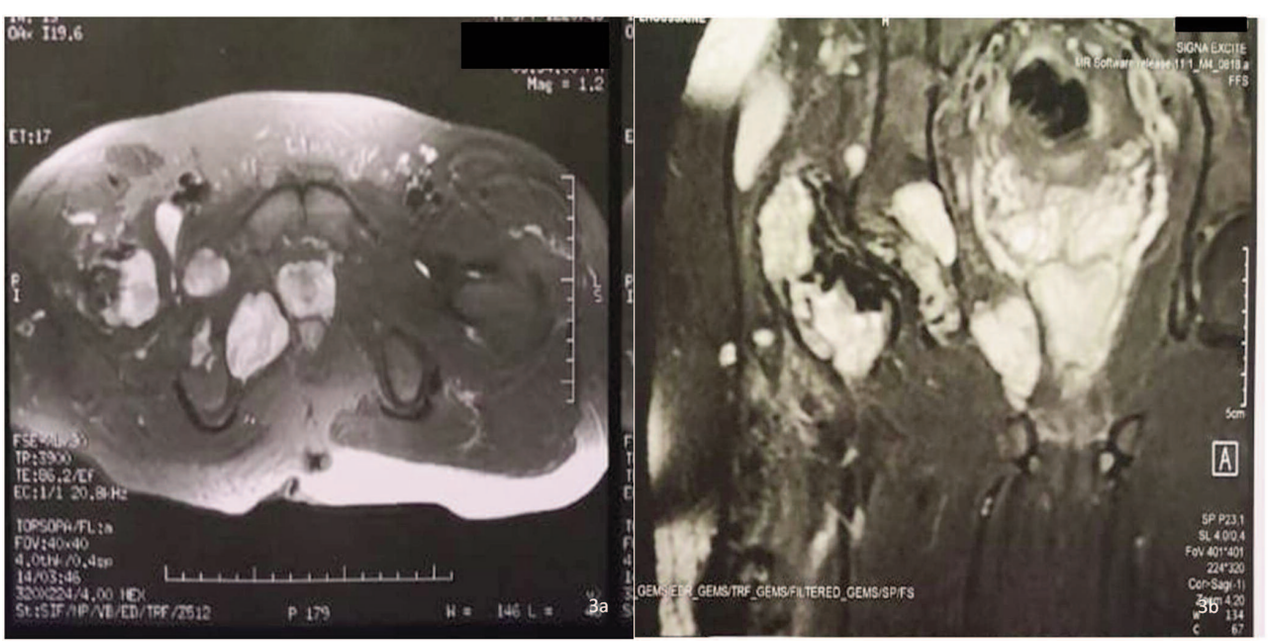

Figure 3. Coronal and axial views of hip and pelvic echinococcosis

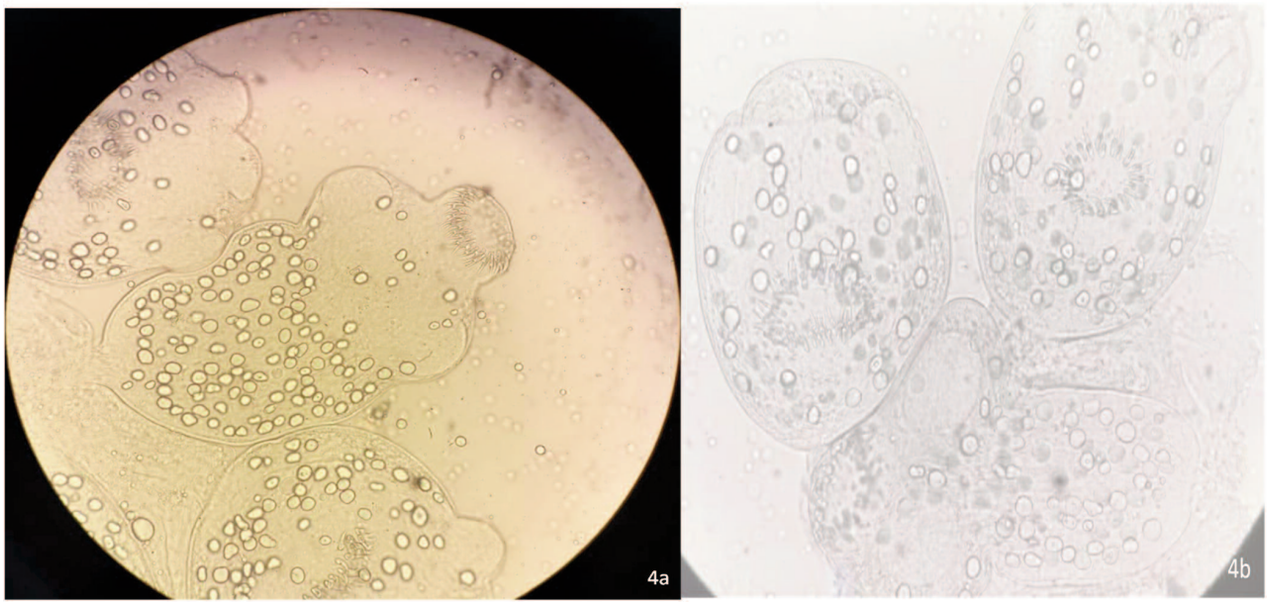

Figure 4. Presence of scolices with dispersed hooklets

tissues [3,11,12].

Diagnosis is usually made postoperatively on cytological study with presence of particulate material containing scolices with dispersed retractile hooklets (PAS and acid fast positive), and bits of the laminated membrane with parallel striations (Fig. 4a,b). Histological study of the taken sample must be systematically carried out in the presence of any atypical radiological manifestation or unexplained pathological fracture.

Surgical management must be carcinological respecting the rules of tumor surgery, with resection of all vesicular formations, which is impossible within extended forms as was the case in our observation. Intra-lesional surgery, frequently source of local recurrence [13] requires the addition of scolicidal procedures with the use of hypertonic saline solutions, formalin, phenol and polymethylmetacrylate [4,9,14,15].

Similarly, osteosynthesis can be performed in the presence of a pathological fracture [4,5,16]. Radical surgery with amputation or disarticulation of the hip as well as hemipelvectomy are rarely used because of the non-acceptance of the majority of patients, and also because of the difficulty and the risks of their realization [17,18].

Medical treatment with Benzimidazole (Albendazole-Mebendazole), antiparasitics with a strong bone and soft tissue tropism, is given sufficiently in the preoperative phase and prolonged in the postoperative period for an average of three months, given the risks of leukopenia and attack hepatic [19]. Sometimes, this medical treatment is the only one used and can be prolonged for several years [14] in situations of an operative contraindication, a difficult access location or very extensive multivesicular forms where carcinological resection is impossible.

A few cases of prolonged remissions have been reported [17,21,22]. In our patient, the medical treatment ensured a remission of 7 years at the cost of a major functional handicap, support and walking are not possible without crutches and the pain is permanent.

Finally, it is important to notice that a recent study about the efficacy and safety of radiotherapy on sheep naturally infected with *E. granulosus* [23], had indicated that this option can be a safe one for treating the disease, especially for patients who do not have opportunities for surgery or are irresponsive to chemotherapy.

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