CLINICAL REPORT

Pelvic bone hydatidosis


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KEYWORDS
Bone hydatid disease; Pelvis; Bone resection; Hip arthrodesis

Summary  Bone hydatid disease is a rare pathology, characterized by its long clinical latency, the absence of radiological specificity as well as its challenging surgical treatment. We report an observation of pelvic bone hydatidosis in a 28-year-old patient, appearing with pain and stiffness of the hip as well as a degenerative aspect on the coxofemoral joint on X-ray. The treatment consisted of an extensive hemipelvectomy with femoropelvic arthrodesis, completed by medical treatment. The functional result was excellent in spite of a break in the fixation material, which allowed a certain mobility at the proximal end of the femur. Hydatid disease of bone is infiltrating, diffuse, slow, and progressive, all characteristics explaining the often-delayed diagnosis. Medical imaging provides accurate analysis for planning a broad surgical resection. The quality of surgical resection is determined according to the risk of recurrence. Pelvic locations are particularly difficult to treat. After an enlarged and difficult surgical resection, reconstruction remains aleatory and poses many technical problems.

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Introduction

Echinococcosis, or hydatid disease, is a parasitosis that in humans is caused by the development of Echinococcus granulosus larvae, which at the adult stage live in the intestines of dogs and other carnivores. Commonly observed in the liver and lungs, echinococcosis of is rare in bone and accounts for only 0.9—2.5% of the manifestations of this disease [1]. Its long clinical latency and the absence of radiological specificity often result in late diagnosis, which contributes to the diffusion and extension of parasitical osteitis [2].

Despite the progress in reconstructive bone surgery and antiparasite medications, treatment remains a challenge [3]. The severity of its prognosis has given it the label of “white cancer” [4]. We report an observation of a case of pelvic bone hydatidosis requiring wide surgical resection and reconstruction by femoropubic and sacral arthrodesis.

Observation

A 28-year-old patient presented in July 2006 for pain and stiffness of the left hip. These symptoms were triggered by a minor injury occurring 9 months before. The patient’s his-
tory revealed an operation at the age of 6 years for a hydatid cyst of the liver.

At the clinical examination, there was no infectious syndrome and the hip was stiff and painful at mobilization. At the left groin and iliac fossa, we observed poorly limited tumefaction, 13 cm in greatest dimension, renitent and only slightly sensitive on palpation. The biological workup showed no particular results: WBC, 7100 cells/mm³, with no hypereosinophilia; sedimentation rate, 15 at the first hour; and C-reactive protein (CRP), 5.6 mg/l.

Standard radiographs (Fig. 1) showed disappearance of joint space in the left hip, a spotted, honeycombed aspect of the periacetabular region of the iliac bone, and a slight cephalic protrusion in the tectal region.

The diagnosis of bone hydatidosis was suggested. Hydatid serology using immunoelectrophoresis was positive. Pelvic CT (Fig. 2) demonstrated the osteitic involvement of the periacetabular region and the wing of the ilium as well as multiple intrapelvic cystic formations. The workup was completed with an MRI (Fig. 3), which eliminated any sacroiliac or sacral involvement and showed the extension of the cystic lesions at the Scarpa triangle. The upper extremity of the femur showed a heterogeneous signal in T1- and T2-weighted sequences, but did not seem to be invaded. In the acetabular area, where cystic images were shown on standard X-rays, the MRI demonstrated a microlacunar aspect with a few centimeter-sized formations on hyperintense T2-weighted images.

The patient had no other hydatid locations and the hepatic cyst operated on in childhood had not recurred. We opted for wide surgical resection. With the patient installed in the right lateral decubitus position, using an enlarged Judet approach, we performed a left hemipelvectomy. From a posterior approach, we released the sacroiliac joint; via an anterior approach, we cut the ipsilateral ilio- and ischiopubic branches. We also resected all the intrapelvic cysts and

Figure 1  Radiograph showing complete disappearance of joint space, cephalic protrusion and honeycomb aspect of the periacetabular region.

Figure 2  CT. Left, periacetabular osteitic involvement. Right, intrapelvic cystic formations (white arrows).
the cysts in the inguinal region. The operative aspect of
the femoral head was reassuring; it was therefore retained.
After having washed it thoroughly with hypertonic physi-
ological solution, we performed the femoropubic arthrodesis.
This was stabilized by directly screwing the femoral head
against the iliopectineal section and particularly by spinal stem
and screw instrumentation between the sacrum and the
superior end of the femur. The ipsilateral iliac crest, macro-
scopically intact, was used for the osseous autograft. We
fitted it and stabilized it with screws against the lateral side
of the sacrum and the trochanteric fossa (Fig. 4).

The postoperative procedure was simple. External
support consisting in a resin pelvic cast was retained
for 3 months. Partial weightbearing was authorized 5
months after surgery. The patient also had medical treat-
ment based on albendazole, following the WHO protocol:
10—15 mg/kg/day, 4 weeks out of 6. This treatment began 2
weeks before surgery, with 10 mg/kg/day, and lasted a total
of 6 months.

Seven months after the intervention, during minor exer-
tion, the patient suddenly felt a crack in the left groin. A
new X-ray showed a fracture in the osteosynthesis material
(stem breakage) and a lysis chamber around the screw at
the iliopubic branch. A new CT was also performed, which
did not show any signs of recurrence.

With this nonunion, we first suggested surgical revision
to change the stem and perform a bone graft. However,
the patient had a satisfactory functional result, he could
walk without a cane, had little pain, and had stable weight-
bearing on one foot. The nonunion area provided a painless
functional range of mobility. No complementary surgery was
undertaken. At 20 months of follow-up, the patient retained
a stable functional result (Fig. 5) and showed no signs of
recurrence. The radiological result was less satisfactory
because of a slight ascension of the superior extremity of the
femur in the pelvis. However, a neo-articulation seemed to
be forming between the trochanteric region and the distal
part of the graft that had consolidated the proximal region
(Fig. 6).

Discussion

Hydatid disease is rife in its endemic state in South Amer-
ica, Australia, but also in the Mediterranean basin and
Central Europe [2]. It is found preferentially in the lungs
(20—30%) and liver (60—70%) [4,5]. Bone involvement is
rare [2,3], accounting for only 0.9—2.5% of all locations
[1]. Bone is infested most often by the hematogenic route
[7], but secondary bone invasion from a primary loca-
tion of the soft tissues is possible [1]. The iliac location
reported herein is certainly primary, with the intrapelvic
and Scarpa triangle cysts secondary to bone involvement.
This is indeed the geographic center of the parasite lesion.
Moreover, no case of iliac bone involvement secondary to
pelvic muscular hydatidosis has been reported in the literature.

In the bones, *E. granulosus* larvae, the parasite responsible for the disease, encounter the mechanical stress of the bone and develop according to a micro- and multivesicular mode by exogenous vesiculation, which weakens the bone over time and destroys it, thus resulting in major lesions [7]. Hydatid osteopathy is infiltrating, diffuse, slow, and progressive, with numerous microvesicles without the parasite encysting [8]. The clinical signs that reveal the disease are not specific and depend on the location. They are dominated by pain and swelling, as in our observation. The notion of contagion and a history of hydatid disease are the signs that orient the diagnosis. The biological tests are also contributive. Hypereosinophilia is inconstant since it is only present in 25% of cases [9] and it is not specific. Immunoelectrophoresis, revealing the arc 5 precipitation characteristic of hydatidosis, is a classical technique, positive in our observation. Currently, we prefer ELISA and Western blot, which are much more sensitive and specific [10]. Standard radiography remains the reference examination for diagnosis. These images most often show poorly limited areolar lytic lesions in the classic honeycomb pattern. There is no periosteum reaction or regional decalcification [2]. Ultrasound can be used to explore the soft tissue in the search for extrasosseous abscesses. It can also diagnose other associated visceral locations [2]. CT provides a complete lesional workup. Bone involvement is seen on hypodense central-osseous images that are unenhanced after contrast injection [11]. Endo- and exopelvic collections are also easily identified. MRI completes CT, providing a better regional workup of the disease, notably in spinal locations [2]. It defines the extent of the bone lesions and adjacent collections with high precision. Images of hydatid vesicles have a hypointense signal on T1-weighted sequences and a hyperintense signal on T2-weighted images [11]. In operated patients, MRI is a good tool for monitoring possible recurrence.

Today’s treatment of bone echinococcosis is medical and surgical [1]. The objectives of medical treatment are reduction of cyst size and sterilization of their contents before and after surgery to treat the small cysts that had not been detected [6,12,13]. Among all the benzimidazoles, albendazole seems to have better digestive absorption [4,14] and its clinical and biological tolerance is good, even for high dosages over prolonged periods [1]. According to the WHO guidelines, albendazole is prescribed at a daily dose of 10–15 mg/kg per day divided into two postprandial doses, in four to six 4-week courses before and after surgery, spaced 2 weeks apart [1]. Regular follow-up of hepatic function is recommended. For our patient, we adopted this therapeutic protocol, but cannot yet draw conclusions on the efficacy of this medical treatment. It should be remembered that the results reported remain controversial and that certain relatively old publications reported bone involvement to be a

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**Figure 5** Clinical aspect at 20 months of follow-up, stable weightbearing on one foot on the operated side, complete extension and 70° active flexion.

**Figure 6** Radiograph at 20 months after surgery. Note the ascension of the upper extremity of the femur and the nonunion between the trochanteric region and the distal part of the graft.
contraindication to treatment with benzimidazole derivatives [2,15]. Surgical treatment aims for complete excision of the hydatid lesions [2]. Recurrence rates after partial excision are very high [4,14]. For our patient, we consider that we obtained total resection of all the diseased bone and the extraosseous abscesses. As for the reconstruction, we opted for femoropubic and sacral arthrodesis. Prosthetic reconstruction was ruled out because the patient was relatively young and active and we deemed it more logical to propose a certain and long-lasting solution. In addition, the risk of hydatid disease recurrence is not insignificant and can threaten the stability of the implant or a possible massive allograft.

The rate of malunion after femoral-ischiatic, -pubic, and/or -sacral arthrodesis is greater than 50% and their functional results are considered insufficient [16]. For this patient, the absence of consolidation is a partial failure in that the patient had stable weight-bearing on one foot and functional range of movement. The relative lack of pain could be explained by secondary denervation at the wide surgical resection.

The follow-up of this observation has been relatively short and hydatid disease recurrence is always possible. Regular and long-term follow-up over time is necessary, best done by CT or if possible MRI [14].

Conclusion

Bone hydatidosis is a rare entity, even in endemic zones. It is often diagnosed late because of its insidious progression. Medical imaging establishes a precise lesional assessment so that wide surgical resection can be planned. The quality of this intervention in terms of risk of recurrence is determinant. Medical treatment seems to be a supplementary contribution whose efficacy has yet to be proven. Pelvic locations are particularly challenging to treat. After wide and challenging surgical resection, reconstruction remains somewhat random and poses a number of technical problems. Early diagnosis and particularly hygiene education in endemic countries remain the best measures that can limit the considerable damage caused by this parasitosis.

Conflicts of interest

None.

References