Recurrence of femoral echinococcosis 5 years after a primary surgical procedure

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Summary
We report a case of recurring femoral hydatid disease in a trisomic-21 patient 5 years after a primary surgery. The patient presented a thigh abscess with a lateral supracondylar area fistula. The workup demonstrated massive osteolysis involving the proximal, diaphyseal, and distal femur as well as multiple soft tissue cystic masses but no metastases. Treatment consisted in cystic masses debridment, extensive bony-curettage of the intraosseous cystic zones, temporary weight-bearing suppression, and albendazole. The patient remains under follow-up and cannot be considered definitely cured.

Introduction
Bone echinococcosis is considered the cancer blanc of the bones [1]. It is rare, even in endemic zones (0.5—2.5% of hydatid disease cases). It progresses slowly and is therefore diagnosed late at the advanced stage of parasitic osteitis. Despite medical and surgical management, it is difficult to treat and its prognosis is not good.

Observation
A 23-year-old trisomy-21 patient was hospitalized on 23 July 2007 in the Infectious Diseases Department of the Nice University Hospital (France) for a right lateral supracondylar fistula that had been progressing for 1 month. For 3 days preceding consultation, he presented proximal tumefaction of the thigh.

The patient’s history included trisomy-21 and morbid obesity (101 kg, 156 cm; BMI, 41.5).
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He had been treated the first time in 2002 for a pathological fracture of the ipsilateral femur with bone hydatid disease (*Echinococcus granulosus*). He was treated with surgery, hypertonic serum, and albendazole (400 mg × 2) with bone union obtained 100 days after injury despite a septic complication requiring early removal of the osteosynthesis material. The albendazole treatment was continued for 22 months and the patient was considered cured [2].

The patient was apyretic. The biological workup found a sedimentation rate at 101 mm, leukocytosis at 5600 WBC/mm³, and CRP at 56.6 mg/l.

The radiographs demonstrated extensive osteolytic lesions of the femur (Fig. 1) and CT showed a voluminous proximal collection (Figs. 2 and 3).

Surgery took place on 14 August 2007. The thigh was opened widely following the fistula trajectory to allow evacuation of the abscess, a cystic cavity measuring 15 cm in diameter. A 1.5-l pseudo-purulent collection containing vesicles was evacuated (Fig. 4). This cavity communicated with the femoral neck via a subtrochlear osteolysis zone. At the fistula loss of lateral cortical substance was also found. Wide curettage of the necrotic tissues and false membranes in both the soft tissues and intramedullary canal was performed with abundant lavage with hypertonic solution and abundant rinsing with isotonic solution.

A single bacteriological sample demonstrated *Staphylococcus epidermidis*, which was considered a contamination.

The parasitological samples demonstrated fragments of anhistic cuticle membrane of *E. granulosus* with no hooks or scolex, suggesting an infertile hydatid cyst.

A second, systematic lavage was carried out on 21st August. Per os albendazole treatment (400 mg × 2) was begun at this time. The postoperative period was without incident and showed no healing problems.
At 3 months of follow-up, the patient was spontaneously without pain, healing was satisfactory, and the last x-rays showed no modification. Weightbearing was not allowed given the risk of fracture.

Discussion

This clinical case illustrates the recurring nature of the disease, as classically described [3,4]. In the preceding observation, the patient walked unassisted 22 months after his initial treatment, with a negative hydatid serology and negative extensive workup [2].

Diagnosis of recurrence is easy in cases with a history of hydatid disease, contrary to the initial diagnosis, which is often made late, whatever the clinical picture: pain, tumefaction, pathological fracture, or even nonunion [5].

Close monitoring of these patients with CT or MRI is necessary because of the risk of recurrence. This was not possible with this patient because of his noncompliance. The literature describes the treatment of femoral hydatid disease as mainly wide resection of the lesions whenever possible, associated with antihelminthic treatment [2,3,6]. Some authors propose reconstructions using a custom-fit mega prosthesis [7] or even disarticulation [8].

In the preceding observation, disarticulation was refused by the patient’s family, his legal guardians. At the time of recurrence, we again opted for conservative treatment.

Preoperative chemotherapy can be proposed when the diagnosis is known in advance [9], which was not the case for this patient.

Femoral diaphyseal fragility remains and the risk of fracture has deferred resuming weightbearing. This patient’s profile does not favor proposing osteosynthesis or mega prosthetic reconstruction.

For the moment we are providing frequent monitoring.

Conclusion

Hydatid bone disease is a particularly serious disease because of its potential for recurrence. Conservative treatment cannot cure the disease. Long-term monitoring is required.

Conflict of interest

None.

References