Hydatid disease of the femoral shaft treated with surgery and hypertonic solution wash-out combined with albendazole

**Kyste hydatique de la diaphyse fémorale traité par l’association chirurgie, sérum hypertonique et albendazole**

M. Winter [1], N. Jacquot [1], T. Balaguer [1], F. De Peretti [1]


**RÉSUMÉ**

Les auteurs rapportent un cas de fracture pathologique survenue dans le cadre d’une hydatidose osseuse localisée à la diaphyse fémorale. Le traitement chirurgical a consisté en un parage large, un lavage au sérum salé hypertonique à 20 %, associé à une ostéosynthèse par clou plaque. Un traitement adjuvant par albendazole a été mené. Les suites opératoires ont été marquées par une rhabdomyolyse et une nécrose locale surinfectée, nécessitant deux réinterventions et l’ablation précoces du matériel. L’évolution locale et générale était satisfaisante à vingt-deux mois. La fracture pathologique a consolidé, aucune localisation secondaire n’a été diagnostiquée et la sérologie hydatique était negative.

L’association chirurgie-sérum hypertonique-albendazole apparaît comme une solution thérapeutique intéressante dans l’hydatidose osseuse.

**Mots clés :** Kyste hydatique, échinococcose, fracture, fémur, albendazole.

**ABSTRACT**

We report a case of pathological fracture of the femoral shaft in a patient presenting a diaphyseal hydatid cyst. Surgical treatment consisted in wide resection followed by washout using a 20% hypertonic saline solution and nail plate fixation. Albendazole was given as adjuvant treatment. Postoperatively, the patient developed rhabdomyolysis with local superinfectected necrosis which required early revision to remove the material. At twenty-two months, the local and general course was satisfactory. The pathological fracture healed and no secondary localization could be identified. Echinococcosis serology remained negative. The surgery-hypertonic solution-albendazole combination appears to be an attractive therapeutic solution for bone hydatid disease.

**Key words:** Hydatid cyst, echinococcosis, fracture, femur, albendazole.
Hydatid cysts are found in a bony location in about 2% of cases. Among these, 30% concern the long bones of the limbs, the femur being affected in about one out of ten cases [Dévé [1], Duran et al. [2], Aslan et al. [3]]. *Echinococcus multilocularis* is generally incriminated as the causal agent [Aslan et al. [3], Dorn et al. [4]]. Also called "white bone cancer" [Dévé [1]] hydatid cyst or echinococcosis of the bone has a poor prognosis. There is a risk of tissue necrosis in the event of a rupture of the cyst or a pathological fracture, leading to diffusion of the parasites into neighboring tissue and general dissemination via the blood stream. Certain authors propose radical surgery with amputation or early disarticulation [Schneppenheim et al. [5], Gorun [6]], others prefer a more conservative attitude [Duran et al. [2]]. Medial treatment with albendazole is generally administered in combination with surgery [Szypryt et al. [7]]. We report a case of hydatid disease which extended to the entire femur and responded to surgical treatment with washout using a hypertonic solution in combination with systemic albendazole.

## CASE REPORT

An 18-year-old male trisomic 21 subject was brought to the Nice University Hospital emergency ward by a local paramedical emergency squad following a fall on August 20, 2002. At admission, the patient presented a spiral fracture of the proximal third of the right femoral shaft. History taking revealed regular contact with dogs and no sojourn in a foreign country in the past. No disease condition related to trisomy 21 was noted.

Preoperatively, the white blood cell count was elevated (14000/mm³) and the eosinophil count was normal.

Radiologically, the initial films exhibited bony remodelling suggestive of a neoplastic lesion. The proximal part of the femur presented heterogeneous confluent zones with no clear signs of cortical remodeling or periosteal reaction (fig. 1).

An emergency transmuscular biopsy was obtained via a lateral open approach, followed by resection and traction. At the emergency procedure numerous membranous elements were seen invading the soft tissue and the femoral shaft. These elements were flat and presented a cerebroid whitish-pearly aspect. They measured 0.5 to 1 cm in width (fig. 2).

The bone and muscle biopsies revealed an invasion by typical hydatid cysts. *E.granulosus* was identified as the causal agent.

A second surgical procedure was undertaken after obtaining the pathology report. A wide lateral approach was used to achieve resection of the intramedullary cysts which had invaded the femoral shaft as well as the trochanter, the neck, and the condyles. The femur and the soft tissues were washed with ten liters of 20% hypertonic solution followed by rinsing with isotonic solution and carcinological debridement. While rinsing with the isotonic solution the color of the muscles changed from red to dark burgundy. The fixation was completed with a plate nail (fig. 3).
Albendazole was administered at the dose of 12.5 mg/kg/d starting as soon as the pathology result was known. Search for extension (hepatic ultrasound, chest x-ray, abdominal, pelvic and cranial CT) did not reveal any extra-osseous localization.

Hydatid serology (ELISA) was 82 UA/ml initially and rose to 355 UA/ml after the second surgery.

The postoperative period was marked by acute rhabdomyolysis and the patient required intensive care for 48 hours with hemofiltration.

Complete necrosis of the wound developed progressively and bacteriological specimens were positive for *Enterobacter* and *Staphylococcus aureus*. The samples were free of any trace of echinococcus.

A third procedure was undertaken fifteen days after the first emergency procedure to remove the necrotic muscular and cutaneous tissue.

A fourth operation was necessary two weeks later for a new debridement and to remove the material which was replaced by transosseous traction. A permanent system was installed to irrigate the tissue with isotonic solution (fig. 4).

After the fourth procedure the postoperative period was uneventful and the clinical and radiological course was satisfactory. Irrigation was removed ten days later.

Biologically, CRP declined rapidly after the fourth procedure. Hydatid serology stabilized at 180-200 UA/ml for 18 months then declined progressively. Serology was negative at 22 months. The thoracic and abdominal images were normal at that time.

Fracture healing was achieved one hundred days after the initial trauma, with 20° lateral rotation and 1 cm shortening (fig. 5).

At 22 months, the patient could walk without assistance, had negative hydatid serology and a negative search for extension. Albendazole was discontinued.

The patient did not present any sign of intolerance (hematologic or hepatic) to albendazole.

**DISCUSSION**

Hydatid disease is a cosmopolite zoonosis affecting numerous mammal species, including humans. The parasite is harbored in its adult form in the gut of the dog which disseminates infesting embrophores into the environment. The parasite is endemic in areas of extensive sheep raising. The preferential localization is the lung and liver with a bony localization being rather rare, but severe. Involvement of the long bones is observed in 30% of the bony localizations with the femur accounting for 10% of cases [Dévé [1], Duran *et al.* [2], Aslan *et al.* [3]].

Locoregional progression of hydatidiasis is a real risk. The progression can involve the entire bone and neighboring soft tissues leading to panosteoechinococcosis described by...
HYDATID DISEASE OF THE FEMORAL SHAFT TREATED WITH SURGERY AND HYPERTONIC SOLUTION WASH-OUT COMBINED WITH ALBENDAZOLE

Constantini. Fracture is frequently the inaugural sign, as in the present case.

Our case illustrates that hydatid disease can spread by manual contamination in southeastern France without travel to an endemic zone. Cases reported in the literature generally involve subjects having resided in North Africa, Turkey, India, or Australia.

The biological diagnosis is based on reliable serology tests [Bonifacino et al. [8]]. Hypereosinophilia is absent in one-third of cases [Gorun [6]].

The initial diagnosis suggested by the first x-ray was probable neoplasia. Diagnostic errors are frequent in the literature [Aslan et al. [3]]. In our case, we retrospectively noted that the cortical bone was thin with no periosteal reaction. The femoral shaft was invaded by round more or less well delimited areas of heterogeneous texture. This aspect was actually not very compatible with a typical osseous neoplasia.

The whitish-pearly membranes observed during the emergency procedure are typical of echinococcosis [Dévé [1], Duran et al. [2], Hooper et al. [9], Gorun [6]].

The initial diagnosis suggested by the first x-ray was probable neoplasia. Diagnostic errors are frequent in the literature [Aslan et al. [3]]. In our case, we retrospectively noted that the cortical bone was thin with no periosteal reaction. The femoral shaft was invaded by round more or less well delimited areas of heterogeneous texture. This aspect was actually not very compatible with a typical osseous neoplasia.

Local treatment by formolization and perioperative radiotherapy appear to be ineffective [Argenson et al. [13], Natarajan et al. [12]]. Rinsing with hypertonic solution [Natarajan et al. [12], Tomak et al. [14]] appears to give good results.

An initial conservative treatment was attempted in the present case because of the age of the patient and the absence of any secondary localization of the hydatid. Surgical treatment with osteosynthesis was associated with carcinological resection of infected tissue. Washout with ten liters of hypertonic solution was probably the cause of the massive tissue necrosis. Rinsing with isotonic solution did not avoid the problem. Massive tissue necrosis with bacterial superinfection required two new procedures for debridement and removal of the implants on day 30 the instauration of transosseous traction and adapted parenteral antibiotic therapy. Permanent irrigation appears to be a satisfactory postoperative solution in this type of situation.

We believe that abundant washing with hypertonic solution enabled complete eradication of the echinococcus but at the cost of massive tissue necrosis. The pathology specimens of the necrotic tissue during subsequent debridements were free of echinococcus.

We can learn two lessons from this case. First the diagnosis of hydatid disease can be established at the emergency procedure on the basis of the presence of white vesicles. This allows immediate precautions in the event of "white bone cancer" [Dévé [1]]. The second lesson is that abundant use of hypertonic solution enabled eradication of the echinococcus despite the fact that the entire femur was infested.

It can thus be suggested that the surgical treatment should include macroscopic debridement of the cysts, prolonged careful impregnation of the operative site using hypertonic solution, comparable to intraperitoneal chemotherapy for digestive cancer. Washing with hypertonic solution must be followed by prolonged washing with isotonic solution and wide debridement of all necrotic tissue which takes on a recognizable deep coloration.

Retrospectively, we regret to have not extended the debridement enough after application of the hypertonic solution.

In light of the pathology results and the clinical course, use of a hypertonic solution is a major element for the treatment of hydatidiasis of the bone. Adjuvant treatment with albendazole (10-15 mg/kg/j) is commonly proposed and should be continued for a long period [Szypryt et al. [7], Natarajan et al. [12], Bonifacino et al. [8], Aslan et al. [3], Agarwal et al. [15]].

We can recall that certain dramatic cases with a pelvic localization must be treated palliatively [Chiboub et al. [16]] with the risk of persistent chronic hydatid fistules.

Regular follow-up with physical examination and standard x-rays as well as monthly hydatid serology can be proposed [Bonifacino et al. [8]]. It is also advisable to search for general dissemination with a thoracoabdominal scan every six months.

CONCLUSION

This case illustrates that the triple association: surgical resection, chemical treatment using prolonged impregna-
tion of hypertonic solution, and drug therapy with albendazole can be proposed for hydatid disease of the bone.

References