

# Hydatid bone disease: a case report and review of the literature

**K Kalinova**

Department of General Surgery, Stara Zagora University Hospital, Bulgaria

**V Proichev**

Department of Orthopaedics, Stara Zagora University Hospital, Bulgaria

**P Stefanova**

Department of Pediatric Surgery, Medical University, Plovdiv, Bulgaria

**K Tokmakova**

Department of Orthopaedics and Traumatology, Medical University, Plovdiv, Bulgaria

**E Poriazova**

Department of Pathology, Medical University, Plovdiv, Bulgaria

---

## ABSTRACT

Hydatid disease may develop in almost any part of the body and can be identified with a combination of clinical history, imaging findings, and serologic results; however, the diagnosis of bone hydatidosis is primarily based on radiographic findings. Bone hydatid disease is often asymptomatic, and its diagnosis is usually made at an advanced stage when lesions have become extensive. We present a case of a 45-year-old woman who was admitted to the University Hospital, Stara Zagora, Bulgaria complaining of pain in her left tibia. Radiographs revealed an oval cyst with a diameter of 3.5 cm, located in the diaphyseal part of the tibia. The cyst was excised, and no recurrence was observed on follow-up. Functional outcome was excellent.

**Key words:** bone and bones; bone cysts

---

## INTRODUCTION

Hydatid disease may develop in almost any part of the body. Bone localisation is rare comprising 0.5% to 2.5% of all human hydatidosis.<sup>1</sup> Although long-term survival is possible, the disease is not easy to eradicate and may be impossible to cure.<sup>2-4</sup> Early diagnosis is uncommon and is primarily based on X-ray findings. Patients usually present at an advanced stage of the disease; therefore, treatment is difficult and recurrence is common.

## CASE REPORT

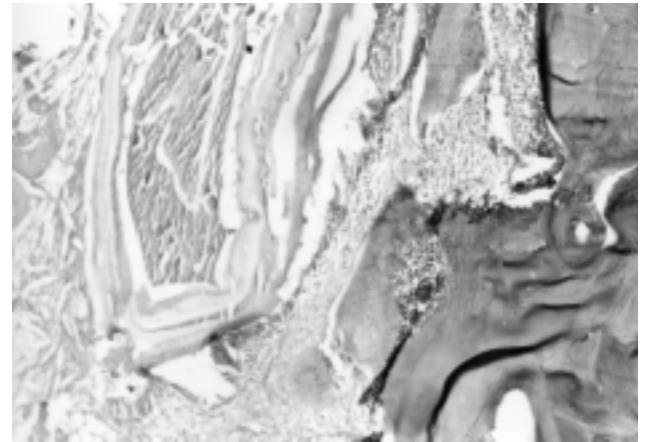
In May 2000, a 45-year-old woman was admitted to the Stara Zagora University Hospital, Bulgaria complaining of pain in her left tibia. Her medical history was unremarkable, and physical examination revealed no abnormal findings. Radiographs revealed an oval cyst with a diameter of 3.5 cm, located in the diaphyseal part of the tibia (Fig. 1).



**Figure 1** Bone hydatid cyst (a) before surgery and (b) 24 months after surgery.

Computed tomographic (CT) scan revealed a hypodense bone lesion without clear boundaries. A periosteal reaction was evident in the cortical area. No significant abnormalities were found on standard blood tests, except for an erythrocyte sedimentation rate of 60 mm/hour.

Surgical exploration of the left tibia revealed a diaphyseal cyst adherent to the surrounding tissues, which were markedly oedematous. Clear fluid was aspirated from the cyst, and the sample was sent for microbiology and serology tests. Povidone-iodine-alcohol solution was injected and cystectomy was performed 10 minutes later. The cyst had a thick wall and an inner layer resembling a germinative membrane. Pathology results revealed hydatid cyst of the tibia. Microscopy confirmed the diagnosis and revealed osseous tissue with hyaline and germinative membranes, lymphocytes, and monocytes (H&E, x200; Fig. 2). The lower extremity was cast and immobilised for 30 days. Oral therapy with albendazole (10 mg/kg/day) was prescribed for 12 weeks. The patient recovered uneventfully and was discharged on postoperative day 10. She was clinically and radiologically followed up for



**Figure 2** Microscopy confirms the diagnosis and reveals osseous tissue with hyaline and germinative membranes, lymphocytes, and monocytes (H&E, x200).

24 months after surgery. No evidence of recurrence was observed, and serological tests were normal.

## DISCUSSION

In 1884, Thomas<sup>5</sup> published a series of 28 cases of hydatid bone disease gathered from isolated reports in the literature. Ivanissevich<sup>6</sup> reviewed 47 cases and published the most detailed work on the condition. More recently, Sapkas et al.<sup>1</sup> reviewed 8 cases of hydatid bone disease in different anatomical locations with follow-up periods from 4 to 16 years, and discussed many related diagnostic and therapeutic problems. The limited publication on hydatid bone disease testifies to the rarity of this condition even in endemic countries.<sup>2,4</sup>

Hydatid bone disease is often asymptomatic, and is therefore usually diagnosed at an advanced stage when lesions have become extensive,<sup>4</sup> as in the present case. The initial location of the lesion in long bones is metaphyseal or epiphyseal, later extending to the diaphysis. Diagnosis is primarily based on findings of X-ray and CT scans. X-ray findings include monolocular, bilocular, or multilocular cysts. Monolocular cysts, as in this case, are rarely observed and are characterised by oval or polycyclic nonspecific lacunae of variable sizes.<sup>4,7,8</sup> Progression of the disease takes place in 2 forms: formation of diverticuli and exogenous vesiculation.<sup>7,8</sup> Potential complications include pathological fracture, infection, and fistulisation of the abscess.

Hydatid bone disease should be considered in the differential diagnosis of osteolytic lesions, especially in endemic areas. Differential diagnoses include chronic osteomyelitis, fibrous dysplasia of bone, osteosarcoma, and benign cystic lesions. The presence of a periosteal reaction, osteosclerosis, and calcification are not specific for hydatid bone disease.<sup>4,9</sup>

The present case of hydatid bone disease was not suspected preoperatively, and immunological tests such as haemagglutination and total and specific immunoglobulin E level measurements were not performed, although they are recommended in the

literature as preoperative diagnostic parameters.<sup>14</sup>

Surgery is the treatment of choice for hydatid bone lesions. Many authors have advocated wide resection of the involved bone along with the surrounding soft tissue as the only definitive treatment of the condition,<sup>3,4</sup> with or without chemotherapy using albendazole or mebendazole. Natarajan et al.<sup>2</sup> emphasised on the need for complete surgical extirpation of the cyst. Curettage and deep radiotherapy have been reported to be insufficient.<sup>10</sup> Despite the limited surgical procedure performed in this case, no recurrence was observed on 24-month follow-up, and the functional outcome was excellent.

## REFERENCES

1. Sapkas GS, Stathakopoulos DP, Babis GC, Tsarouchas JK. Hydatid disease of bones and joints. 8 cases followed for 4-16 years. *Acta Orthop Scand* 1998;69:89-94.
2. Natarajan MV, Kumar AK, Sivaseelam A, Iyakutty P, Raja M, Rajagopal TS. Using a custom mega prosthesis to treat hydatidosis of bone: a report of 3 cases. *J Orthop Surg (Hong Kong)* 2002;10:203-5.
3. Mnamneh W, Yacoubian V, Bikhazi K. Hydatidosis of the pelvic girdle—treatment by partial pelvectomy. A case report. *J Bone Joint Surg Am* 1977;59:538-40.
4. Zliti M, Ezzaouia K, Lebib H, Karray M, Kooli M, Mestiri M. Hydatid cyst of bone: diagnosis and treatment. *World J Surg* 2001;25:75-82.
5. Thomas JD. Hydatid disease. Adelaide: E Spiller; 1884:47-9.
6. Ivanishevich O. Hidatidosis osea [in Spanish]. Buenos Aires: Amorrortu; 1934:16-8.
7. Markakis P, Markaki S, Prevedorou D, Bouropoulou V. Echinococcosis of bone: clinico-laboratory findings and differential diagnostic problems. *Arch Anat Cytol Pathol* 1990;38:92-4.
8. Saidi F. Hydatid cysts of bone. In: Saidi F, editor. *Surgery of hydatid disease*. 1st ed. London: WB Saunders; 1976:135-8.
9. Torricelli P, Martinelli C, Biagini R, Ruggieri P, De Cristofaro R. Radiographic and computed tomographic findings in hydatid disease of bone. *Skeletal Radiol* 1990;19:435-9.
10. Engin G, Acunas B, Rozanes I, Acunas G. Hydatid disease with unusual localization. *Eur Radiol* 2000;10:1904-12.