A 48-Year-Old Somali Woman with Hip Pain

(See pages 764–5 for the Photo Quiz)

Figure 1. Noncontrast computed tomography scan of the pelvis. The arrow points to a lytic lesion in the posterior mid-right ilium characteristic of a space-occupying hydatid cyst.

Diagnosis: hydatid cyst bone disease.

Cysts removed from the iliac bone cavity (figures 1 and 2) were consistent with hydatid cysts due to Echinococcus species. Microscopic examination confirmed the diagnostic features of cystic hydatid disease, including cysts surrounded by prominent laminated acellular layers. Shown is a characteristic cyst with an inner germinal layer and protoscolex with hooklets (figure 3). The fluid and protoscoleces contained within the cysts are known collectively as hydatid sand. Both macroscopic and microscopic features were most consistent with cystic hydatid disease.

The cavity was completely curetted and irrigated with several liters of saline before the wound was closed. To support the histological diagnosis, serological testing was performed, and results were positive for immunoglobulin G–class antibodies to Echinococcus species by enzyme-linked immunoassay. Computed tomography (CT) of the chest, abdomen, and pelvis did not show evidence of multifocal disease. The patient initiated therapy with albendazole (200 mg twice per day) with the plan to continue therapy for 8 months. Follow-up CT scans of her pelvis at 1 and 8 months after hospital discharge showed the lytic lesion to be stable. The patient continued to improve and ultimately discontinued albendazole after 8 months of therapy.

Hydatid disease is caused by the larval forms of Echinococcus cestodes. The most common species are Echinococcus granulosus and Echinococcus multilocularis, which cause cystic and alveolar hydatid disease, respectively. The geographic distribution of these 2 species differ markedly, with E. granulosus having a worldwide distribution and E. multilocularis being restricted to temperate regions of North America, Europe, and Asia [1]. In sub-Saharan Africa, especially in East and West Africa, cystic hydatid disease is highly endemic [2]. The presence of the laminated acellular layer surrounding the cyst in figure 2, as well as the recent emigration of our patient from an area of endemcity [3], supports a diagnosis of E. granulosus cystic hydatid disease. A laminated layer is not seen surrounding the...
Figure 2. Cystic structures removed from the pelvic lesion. Note the characteristic variation in size and shape of the cysts. Many of the cystic structures have been cut open to reveal the mostly smooth inner lining.

cysts associated with *E. multilocularis* alveolar hydatid disease, which are instead separated by cellular connective tissue [4]. The definitive hosts (which harbor adult tapeworms) are carnivores (mainly dogs and foxes), and the intermediate hosts include sheep, pigs, and cattle. Humans are accidental hosts and become infected by ingesting *Echinococcus* eggs that are present in water or food contaminated with fecal material from the definitive hosts. In the intestine, the echinococcal oncospheres penetrate the mucosal membrane and disseminate through the portal blood vessels to the liver, which is the pri-

Figure 3. Tissue section of pelvic material from the open biopsy sample (hematoxylin and eosin stain; original magnification, ×100). The diagnostic features of cystic hydatid disease, including a cyst surrounded by a laminated layer (LL), and lined by a germinal membrane (GM), are shown. Within the cyst is a protoscolex with prominent hooklets (inset; original magnification, ×400). The LL is characteristic for *Echinococcus granulosus* cystic hydatid disease and is not seen with *Echinococcus multilocularis* alveolar hydatid disease.
mary and only site of infection in 75%–80% of cases. The second most common site of infection is the lungs (15%–25% of cases), but other organs can also become infected, including the brain, skeletal muscle, spleen, and bones [5, 6]. Bone is a rare site of infection for all types of echinococcal disease, accounting for 0.5%–4% of cases [6–9]. The most common site for bone infection is the spine (∼50% of cases), followed by the pelvis and hip joint [5, 10]. There is no consensus on the frequency of primary hydatid bone disease (ie, without liver or lung involvement) [7, 8, 11–13]. In our patient, no additional skeletal cysts were detected, suggesting that she had a primary infection of the bone. Growth of the hydatid cysts in bone is slow but destructive, because no adventitia is formed. Cysts are free to multiply, and because of the rigid nature of the bone tissues, they tend to invade the trabeculae and the medullary cavities [6, 7, 10, 14]. The diagnosis of hydatid cyst bone disease is usually delayed, because patients are often asymptomatic until the late stages of the disease and may present with nonspecific symptoms, including pain and edema [8, 13, 15].

Making a preoperative diagnosis of hydatid bone disease is difficult, especially in areas of low endemicity. The differential diagnosis for our patient, based on imaging studies, included chondromyxoid fibroma, chondrosarcoma, osteosarcoma, or a microbial abscess. Hydatid disease was not expected until the contents of the bone lesion were visualized. According to studies reported in the literature, other conditions that may be included in the differential diagnosis include plasmacytoma, fibrous dysplasia, giant cell tumor, and lymphoma [6].

Treatment usually consists of a combination of surgery to remove the cyst and chemotherapy (specifically, albendazole therapy) [7, 16]. The World Health Organization recommends a dose of 10–14 mg/kg/day for 4–6 weeks [8]. However, a review of the literature suggests that treatment decisions are often made on a case-by-case basis and are guided by the severity of the infection, the location of the cysts, and the patient’s tolerance of the drugs [8, 15, 17, 18]. Alveolar hydatid disease often requires more aggressive surgical and chemotherapeutic management. Recurrence of hydatid bone disease ranges from 30% to 50% of cases [8, 10, 15] and can occur up to 45 years after the original diagnosis and surgery [5, 15].

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References


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