Multilocated hydatid cysts involving the liver, the spleen and the scapula

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INTRODUCTION

This article reports an unusual case of hydatid disease involving the liver, the spleen and the scapula to stress the importance of maintenance of both a high index of suspicion and clinical awareness for making an early diagnosis and initiating the proper therapeutic management. Recognition of hydatid disease is not easy, because of its rarity and the vagueness of its symptoms, and as such the diagnosis may be missed or delayed. This case emphasizes the importance of considering hydatid disease, an almost forgotten cause of cystic lesions in most developed countries, in the differential diagnosis of cystic and/or lytic lesions located in multiple sites.

DISCUSSION

Hydatid disease is medically and socioeconomically the most serious tapeworm infection (Scully et al, 1979). Although North and South Africa, Middle East, South America, Central Europe, the Mediterranean, Australia and Canada are the main regions of endemism, physicians should always be alert because of the mobile state of the world's population. The liver is the most commonly affected organ, with right lobe involvement seen in 80% of hydatid disease cases (Monroe, 1985). Eosinophilia and discomfort are present in 55% and about 50% of the cases respectively (Monroe, 1985). Marginal calcification of an echinococcal cyst, which makes the cyst evident on a plain radiograph, usually occurs after 5–10 years of infection in 20–30% of patients (Czerniak et al, 1964). Splenic echinococcosis is uncommon, representing less than 2% of hydatid disease (Bonakdarpour, 1967).

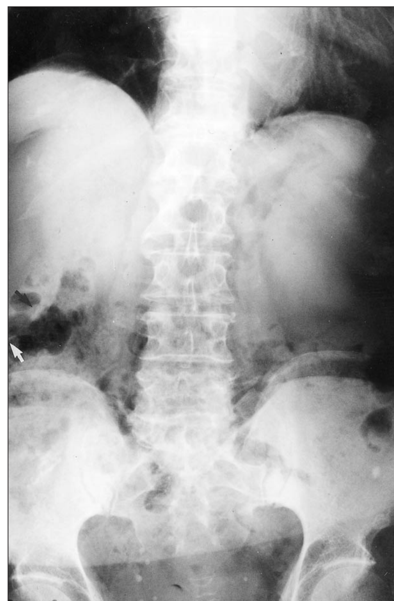
CASE REPORT

A 75-year-old healthy woman presented with mild, right hypochondral discomfort and heaviness present for at least 2 days before admission to the hospital. She also complained of pain in the region of her left shoulder, which was moderate in nature and of approximately 1 year's duration. Shoulder pain had led to the limited use of her left arm. The patient had a history of intimate contact with sheep and dogs. Clinical examination revealed a tender, enlarged liver (5 cm below the right costal margin), as well as a tender scapula on palpation. Laboratory tests showed the white blood cell count to be 7000/mm^3 with 8% eosinophils. The erythrocyte sedimentation rate was 30 mm per hour. Serological tests for *Echinococcus granulosus*, which included immunoelectrophoresis and indirect immunohaemagglutination assay, were strongly positive (IHA 1/800).

A plain abdominal radiograph showed the enlarged hepatic shadow and a curvilinear calcification lying within it (*Figure 1*). An ultrasound study was then performed to further characterize the lesion. A hyperechoic band located in the right lobe of the liver associated with an acoustical shadow representing dense calcification was found (*Figure 2*). Two roundish, hypoechoic areas located in the enlarged spleen were found incidentally during the ultrasound examination of the abdomen (*Figure 3*). The diagnosis of splenic hydatid disease was favoured because of the simultaneous presence of the cystic lesion in the liver. The roentgenographic examination of the left scapula showed multiple, variously sized, round or oval osteolytic lesions, giving a honeycomb appearance of the inferior part of the scapula (*Figure 4*).

On the basis of clinicoradiological features and laboratory findings a diagnosis of hydatid disease was proposed. A thorough clinical, radiological and ultrasonographic search did not reveal any other sites of hydatid cysts. The patient proceeded to surgery where she underwent local hepatic resection, splenectomy and local total excision with a wide healthy margin in the scapula. Histological examination of the material excised from the liver, the spleen and the scapula confirmed the diagnosis of hydatid disease. The patient made an uneventful postoperative recovery and was discharged 1 week later. Pain subsided totally following surgery. At follow-up examination 6 and 12 months later, the patient remained asymptomatic, and had no clinical or radiological evidence of the disease in the primary sites or elsewhere.

Figure 1. Frontal radiograph of the abdomen (supine position) shows focal calcification in the hepatic parenchyma (arrows).



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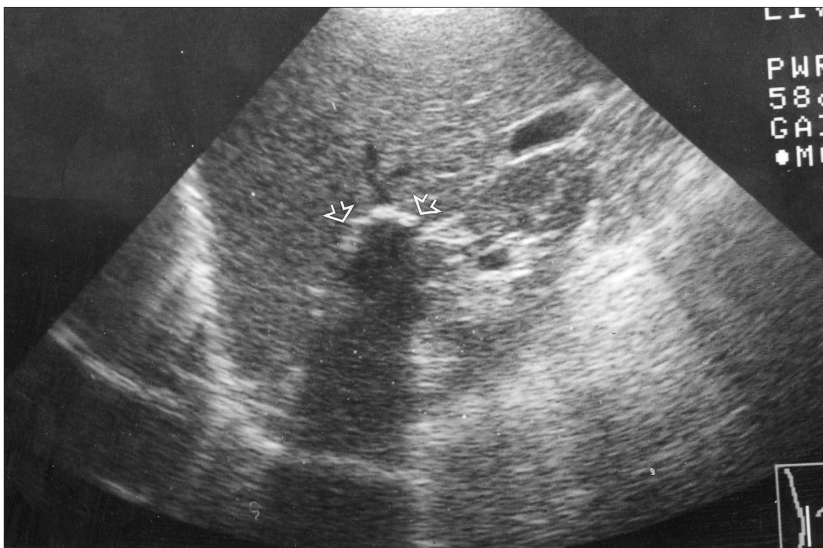

Figure 2. Ultrasound of the liver shows a hyperechoic lesion located in the right lobe (arrows), with an associated intense acoustical shadowing cone caused by calcification.

It is interesting that hydatid disease of bone occurs in only about 1% of all hydatid disease cases, possibly because of the mechanical resistance bone offers to the growth of parasites (Alldred and Nisbet, 1964). Skeletal hydatid disease is always primary, as it is not spread from other sites (Hooper and McLean, 1977). The most commonly affected skeletal sites are the spine, the pelvis and the hip joint (60% of cases). The ribs and the scapula are reportedly involved in 8% of the cases (Bonakdarpour et al, 1973).

Diagnosis of hydatid disease may be particularly perplexing, because of the lack of clear clinical manifestations of the disease, the relatively silent and obscure course, and the difficulty in interpreting laboratory findings. Following the trend of immunological findings is invaluable, because when they become negative (as in the event of total calcification) 6–18 months postoperatively there is only a small chance of recurrence, whereas when they remain positive up to 24 months after surgery there is a high possibility of the presence of persistent cysts (Cardona et al, 1983). The therapeutic difficulties of hydatid disease are the outcome of the behaviour of the parasite, which is comparable to that of a malignant neoplasm as it is characterized by local invasion, high rate of local recurrence, and other complications such as cyst rupture, fistulae and fractures. HM

Figure 3. Ultrasound of the spleen reveals two discrete, roundish hypoechoic areas (arrows), separated by normal parenchyma. Splenomegaly is also present.

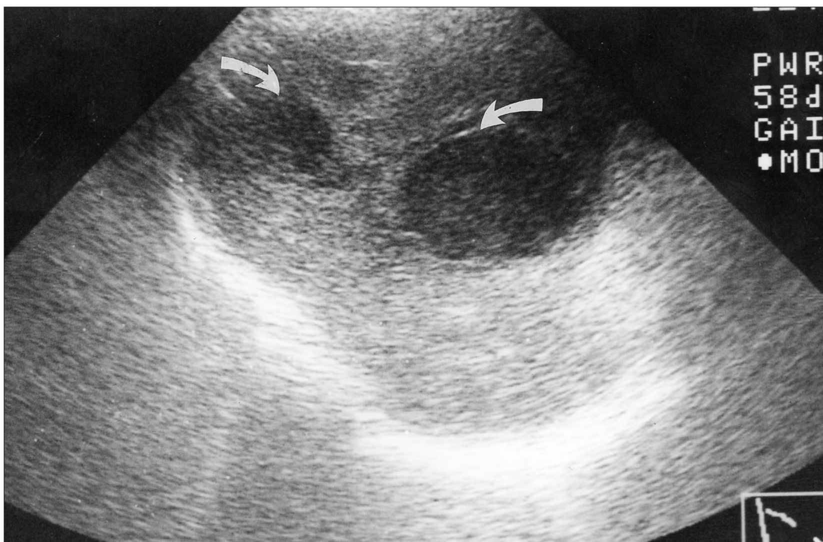

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Figure 4. Lateral radiograph of the left scapula shows well-defined, multilocated osteolytic lesions, accompanied by expansion and thinning of the cortex in the inferior one-third of the scapula. No adjacent soft tissue density or swelling is present.