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## Echinococcosis of the Liver

### Report of a Case

Roberto Benedetti, M.D.\* and Joseph L. Ponka, M.D.\*\*

*Echinococcosis of the liver is so rare in our practice that the case presented is the only one documented in the records of Henry Ford Hospital. The massive enlargement of the liver required surgical intervention. Marsupialization was the treatment of choice. In spite of post-operative complications, the patient made a satisfactory recovery.*

Echinococcosis is an uncommon disease in our practice and, when seen, it is likely that the disease was acquired in a foreign country. The case presented here illustrates problems encountered in diagnosis and surgical management, including management of the post-operative complications of infection and biliary fistula.

#### Case Presentation

The patient, a native of Iraq who had resided in this country only two years, was first seen at the Henry Ford Hospital in 1961 at the age of 70. Her chief complaints were rectal bleeding and abdominal discomfort with mild pain related to the ingestion of food. She reported no vomiting.

Physical examination showed an obese and hypertensive woman with a large, firm, irregular, non-tender mass in the right upper quadrant. This appeared to be contiguous with the liver.

The clinical impressions included hepatoma or adenocarcinoma, metastatic to the liver; the possibility of echinococcosis; also benign liver cyst, and hemangioma or hydrops of the gallbladder.

Her hemoglobin was 11.2 gm/100 cc, the

leucocyte count was 10,500/cu mm with a normal differential. Bromsulphalein (BSP) retention was 8%. Stool examination revealed the presence of pinworms.

The serum was positive for hydatid antibodies by the complement fixation test (titre 1:128) and hemagglutination test (titre 1:5260). The Casoni skin test was positive.

X-ray examination of the abdomen revealed a large, soft tissue density which extended to the level of the iliac crest and could not be separated from the hepatic area (Fig 1). Renal and psoas shadows were normal. The radiologist suspected hemangioma. Upper gastrointestinal films revealed antral ulceration. Barium enema examination, cholecystograms, and intravenous pyelograms were also negative.

The patient was given the diagnoses of hydatid cyst, peptic ulcer, obesity, and hypertension, and then placed on an ulcer regimen and weight reducing diet. While she was being followed in the out-patient department, the cyst gradually enlarged and she developed a mild anemia, the hemoglobin dropping to 10 grams. Surgery was not advised, because of her age.

In 1969, eight years after her first admission the cyst enlarged enormously. Her abdominal pain became continuous, severe and intolerable.

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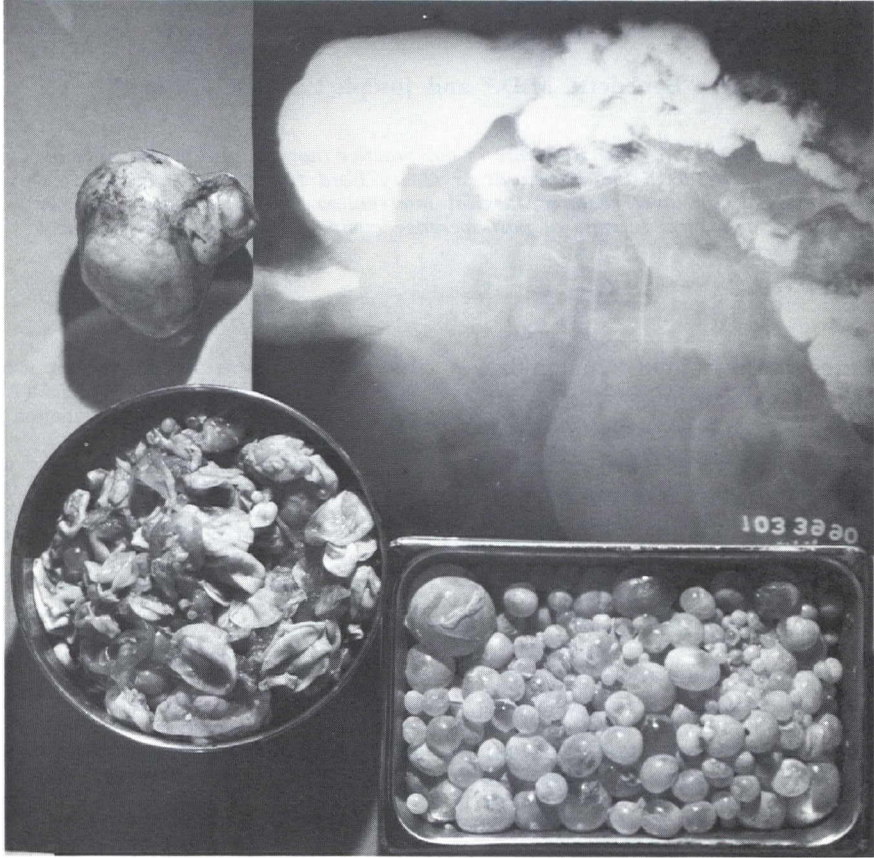


Figure 1

Upper gastrointestinal studies show the tremendous displacement of the stomach, colon and small intestine downward towards the pelvis and towards the left half of the abdomen. The multitude of cysts are shown in two basins.

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Operative treatment was advised as the only solution.

### Operative Treatment

The operation was performed May 7, 1969 under catheter spinal anesthesia. Following a right subcostal skin incision, a large circumscribed mass was found imbedded in the round ligament, but was not attached to the liver itself. With sharp and blunt dissection, this cyst was enucleated from the surrounding tissue. Bleeding vessels were clamped and ligated with plain catgut sutures. Two small individual cysts, attached to the liver by thin pedicles, were also removed intact by clamping, dividing and ligating the pedicles. The massive right lobe of the liver was firm to palpation and reddish brown in color. No discernable normal liver tissue was present on the anterior surface of the liver. Only a small crescent of normal appearing liver was present in the anterior portion of the left lobe. Firm abnormal tissue dominated elsewhere. Because of the great size of the liver and the danger of rupture of Echinococcal cysts, only limited exploration of the lower abdomen was carried out. Throughout the operation, extreme care was taken to prevent spillage of cyst contents. The right lobe was isolated from the wound with multiple thicknesses of laparotomy pads and a large caliber needle was introduced into the white fibrous area. Only a small amount of brownish green fluid was removed, and even after adjustment of the needle, no further fluid was obtained. Several cubic centimeters of 2% Formalin solution were injected but could not be withdrawn. Therefore, it was felt necessary to enter the cyst cavity.

After carefully isolating the gigantic cyst from the wound and peritoneal cavity, it was incised. Approximately 1500 daughter cysts were removed with an ordinary tablespoon as an effective instrument (Fig 2). After evacuation was complete, it became apparent that the entire right lobe of the liver had been replaced by cysts (Fig 2) and all that remained was a thin outer layer of liver tissue with cyst wall. The gallbladder was found lying on the inferior aspect of the cyst wall.

The cyst also extended posteriorly toward the left lobe of the liver. A large portion of the cyst wall was removed (Fig 3). The inside wall of the cyst was then scraped and the wall swabbed with full strength Formalin. The adventitia of the cyst was disturbed very little. A small amount of bile could be seen escaping from the wall here and there. The packs were then carefully changed and the edges of the cyst swabbed with Formalin.

After the abdominal wall was closed, the cystic opening was sutured to the peritoneum and anterior abdominal wall with interrupted chromic catgut sutures. Marsupialization was elected as the procedure of choice to handle the problem of drainage. Four Penrose drains and a sump were placed through the opening and the skin was closed around the opening.

The pathologist reported that the infestation was due to *Echinococcus granulosus*.

Although the patient tolerated the

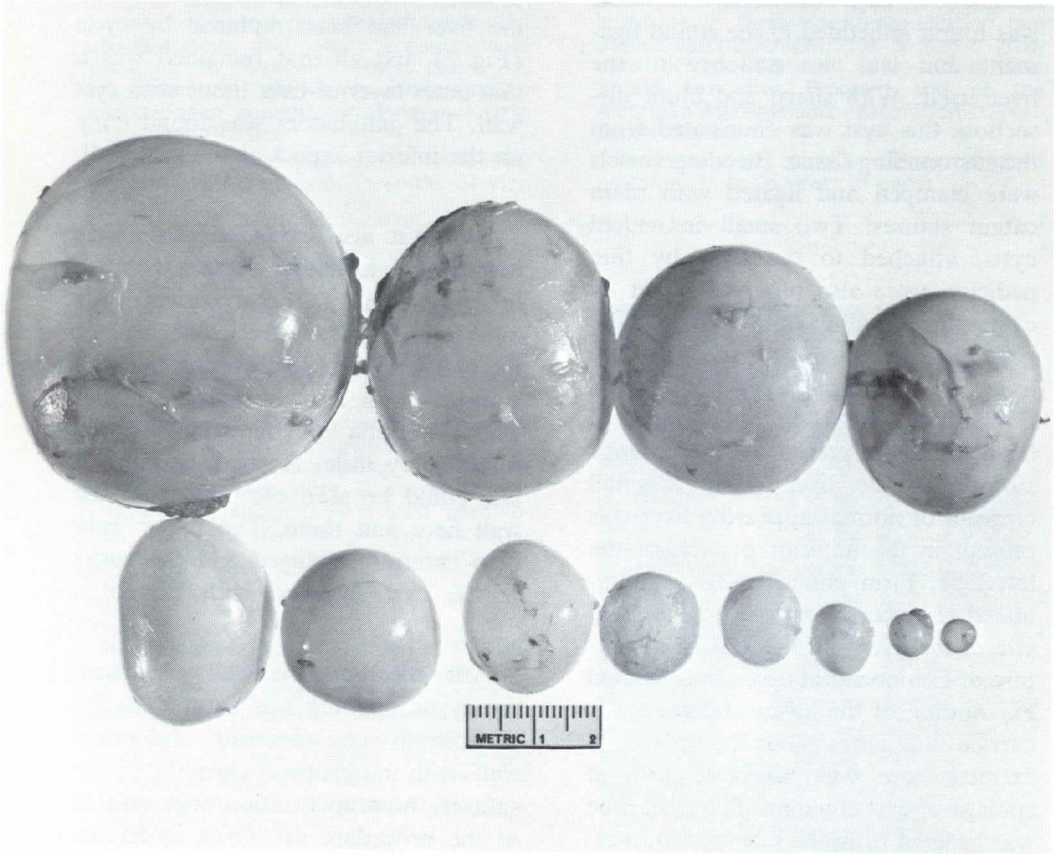


Figure 2

Photograph illustrates the great variation in size of the cysts.

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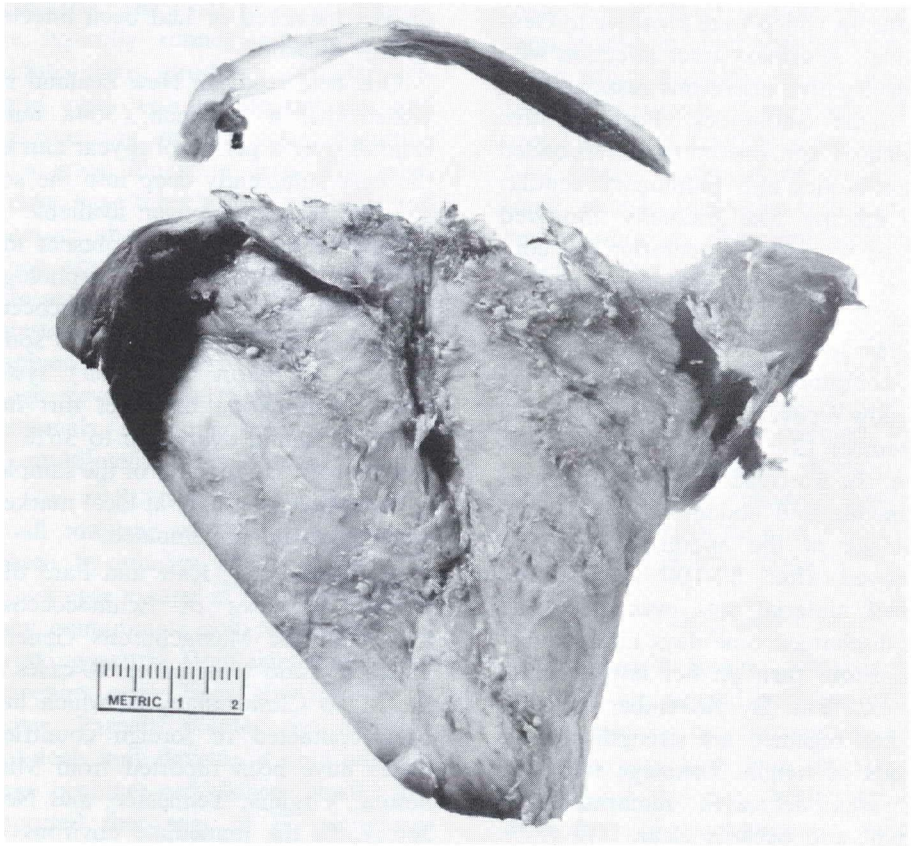


Figure 3

Photograph shows characteristics of the dense capsule and small strip shows its thickness.

procedure satisfactorily, her post-operative course was stormy. Daily drainage from the marsupialized cyst amounted to 200 to 400 cc of sanguineous, bile-stained fluid. The temperature varied from 99.5 F (37.5 C) to 102.2 F (39 C). Wound culture revealed two strains of pseudomonas organisms which were sensitive to Gentamicin. A urinary tract infection with gram negative organisms responded to the same antibiotics. Post-operative atelectasis and pneumonitis responded to antibiotics and pulmonary ventilatory therapy. She gradually improved and six weeks after operation her condition justified transfer to a nursing home.

Drainage from the marsupialized cyst continued to be a problem during her slow recovery as an outpatient. On September 12, three months after operation, she was readmitted for treatment of anemia and abdominal discomfort. Drainage of the operative area was improved after 80-100 cc of bile-stained material was evacuated. She was discharged nine days after admission. From then on her improvement was excellent. By November of 1969 she had regained her strength and 19 pounds of weight. Drainage from the cyst area decreased remarkably in amount and became clear.

#### Epidemiology

Echinococcosis is a cyclozoonotic infection of world-wide importance today because of the rapid movement of people from one continent to another. The adaptability of these parasites to a wide variety of host species has made possible the broad distribution of the infection from the Arctic Circle to the tropics. It is prevalent in sheep and

cattle raising countries. Sir William Osler reported the presence of *Echinococcus granulosus* in Canada in 1883.<sup>1</sup> The disease is endemic in the Northern territories where approximately 10% of the Indian population of British Columbia, and 40% of Eskimos and Indians in the Yukon district harbored or had been infected by this parasite.<sup>1,2</sup>

One field study in New Zealand revealed that a 120-inch (3048 mm) rainfall over a period of a year carried the eggs sufficiently deep into the soil so they were no longer available to grazing animals.<sup>3</sup> It is at present impossible to differentiate morphologically between eggs of *Echinococcus* and other taeniid tapeworms. In some sections of Beirut (Lebanon) systematic examination of street dirt has revealed taeniid ova in up to 36% of the samples. Up to 34% of the samples of salad greens bought at local markets were similarly contaminated.<sup>5</sup>

In this country, Katz and Pan<sup>4</sup> discussed 74 cases of Echinococcosis treated at the Massachusetts General Hospital; Judd reported on 90 cases at the Mayo Clinic, many of which had been contacted in foreign countries. Cases have been reported from Minnesota, Virginia, Tennessee, and New Jersey. In the immediate environs of Salt Lake City, where the disease is endemic, 19 cases have been reported, all confirmed in surgery. In only one of these cases was the disease contacted outside of Utah.<sup>2</sup> The prevalence of dog infestation is a good index of local area infection and degree of risk to man.

There were 96 cases of hydatid cyst of the thyroid described in the intestinal literature up to 1950.<sup>6</sup> According

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to Katz and Pan<sup>4</sup> 17 cases of hydatid cyst of the spleen were reported in the United States from 1882 to 1956.

### Pathology

Echinococcosis is a disease caused by the parasitic tapeworms *Echinococcus granulosus* and *Echinococcus multilocularis*. The cysts of *E. granulosus* are typically round and benign. *E. multilocularis* produces alveolar hydatid cysts with locally invasive and metastasizing microvesicles, resulting in a grave illness in man. The tapeworms have three to five segments and measure about one-fourth of an inch. They are host specific. Development into sexual maturity occurs only in the intestinal tract of carnivores, usually canines. In contrast, the cystic larval stage develops in a wide variety of mammals (*E. granulosus* in herbivores, *E. multilocularis* in rodents).

The adult tapeworm lives in the small intestine of the definitive host where it attaches itself to the villi. When eggs escape in the canine feces, they contaminate soil, vegetation and drinking water. High winds may also transport eggs. Eggs sink rapidly in water. Parnell<sup>5</sup> pointed out that hydatidosis may develop if only one embryo becomes established. Nosik<sup>5</sup> first reported that eggs of *E. granulosus* were resistant to Formol, and further data involving a wide range of substances revealed that no compound can effectively kill the eggs. They are susceptible to desiccation and they are rendered harmless by boiling water; however, infective material should be boiled for at least five minutes.<sup>5</sup>

The capsule of the ingested egg is dissolved by the gastric juice and the larva migrates through the intestinal

wall and reaches the liver via the portal system. Cameron estimated that 70% become located in the right lobe.<sup>7</sup> The remaining larvae are carried to the lungs, muscle, bone, kidney, spleen, brain, heart, thyroid, etc., where they may become encysted. By the end of the fifth month the cyst presents the characteristic outer hyaline protective capsule, and an inner geminative membrane which produces brood capsules and daughter cysts. A number of these brood capsules become detached and float freely in the cystic fluid. The small, sand-like particles with a granular sensation to palpation have been called "hyatid sand." Occasionally, the cyst will produce brood capsules but no scolices and is called an acephalocyst. Scolices may be liberated from the mother cyst as a result of rupture, leakage, or surgical intervention. Many of them die, but others develop into new hydatids; the resulting condition carries poor prognosis and is known as "secondary Echinococcosis."

### Diagnosis

Symptoms in Echinococcosis are the result of mechanical pressure exerted by the cyst on the surrounding tissue and thus vary both with the size and location of the cyst. Moderate sized cysts may be asymptomatic, or may produce a sensation of heaviness in the abdomen. Large cysts may cause mechanical obstruction. Hepatomegaly may be found on physical examination. Jaundice may ensue from pressure on major biliary ducts.

In 1924 Deve presented a triad of symptoms in this disease: (a) the teenager with biliary colic, (b) jaundice, and (c) laminated membranes in feces.<sup>8</sup>



Eosinophilia, usually 5% or more, is present in over half of the patients.

Casoni intradermal test is reliable in approximately 85% of patients. The test is performed by intradermal injection of 0.25 cc of the antigen, which consists of hydatid fluid from fertile cysts of animal origin. In a positive reaction the patient develops a raised wheal with surrounding erythema, at least one inch in its smallest diameter. Negative reactions are any smaller than  $\frac{3}{4}$  of an inch in diameter. False negative reactions are not uncommon in children with lung cysts, since the infected cysts present a negative Casoni test.

Serological tests presented by Fischman<sup>9</sup> include:

1. Whole-scolex complement fixation test, positive in 92.3% of cases.
2. Hydatid complement fixation test, positive in 73.8%.
3. Latex-agglutination test, positive in 87.3% (titre above 1:400).
4. Scolex extract latex test, positive in 80%.
5. Fluorescent antibody test, positive in 81.5%.

A whole-scolex complement fixation test and one agglutination (latex) test are suitable for routine purposes.<sup>9</sup>

X-ray films of the abdomen are helpful in the diagnosis. In Australia, reticular calcification of the entire cyst is considered the most specific x-ray sign and is almost diagnostic of hydatid disease.<sup>10</sup> Intravenous cholangiograms will show distortion of the biliary ducts by cysts greater than 5 cm. Liver scanning may be very helpful in localizing the cyst. Selective arteriography of the celiac axis and superior mesenteric artery distinguishes the hydatid cyst

from other causes of avascular areas in the liver through distribution of contrast medium between the cyst wall and pericystic layer in the venous hepatogram phase.<sup>11</sup>

#### Principles of Treatment

Chemotherapy for patients with hydatidosis is a complex problem. Cuervo Garcia proposed a treatment of 3 cc of 50% solution of thymol together with 1% iodine in oil. This preparation is administered intramuscularly in series of 15 injections on alternate days. Three series of injections are given with 10-day intervals between the series.<sup>12</sup> *In vitro* activity was demonstrated for Gentian violet, tartar emetic, emetine and acriflavin. Scolicides destroy the larva, brood capsules and germinal epithelium through dessication, but Hicken et al<sup>2</sup> point out that chemical sterilization is not always complete. Some cysts continue to perpetuate their larval infestation.

Effective chemical scolicides at the time of surgery are: hydrogen peroxide, cetrimonium bromide, iodine, octoglycin, dihydropentiodide, sodium hypochlorite, glycerol, and Formal. Of these, Formal and sodium hypochlorite are the most available ones.

Stibophen, diethylcarbamazine, tetracycline and oxytetracycline were inactive. Promising results have been recently obtained *in vitro* with niclosamide, bithionol, and dichlorophen.<sup>13</sup>

In spite of efforts at developing effective drug therapy, at the present time complete surgical excision offers the best possibility of eradicating the disease.

The surgical approach depends on the location of the cysts. Those located in the upper surface of the liver are best approached posteriorly by resect-

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ing the 10th and 11th rib. The pleura should not be opened. The cyst must be kept well isolated from the surrounding operative field. If needles are used for aspiration, utmost care must be taken not to spill fluid. After evacuation of the cyst content, Formalin 1.5% solution may be used, or 30% saline solution as recommended by Fontant in 1961.<sup>8</sup> No attempt should be made to remove the adventitia of the cyst because of the frequent presence of vessels and biliary ducts adjacent to it.

When cysts rupture into the biliary tract, operative cholangiography is considered essential to locate accurately the mother cyst and to assure that the common bile duct has been cleared of daughter cysts. If biliary flow is free from the cyst wall, the use of cystojejunostomy using a Roux-en-Y loop must be considered. Internal drainage is preferable to external drainage.<sup>8,14</sup>

It is generally agreed that excision of the cyst is preferred treatment whenever possible. If it is necessary to enter the cyst, spillage of the contents must be avoided if secondary implantation is to be avoided. Injury to the adventitia is hazardous since it may result

in hemorrhage and large biliary fistulae.

Marsupialization is indicated in cases where the surgeon feels that this is the most effective way of evacuating and draining the cyst.

Finally, blind needle aspiration of cysts through the intact abdominal wall is not recommended because of the danger of spillage and secondary implantation. Furthermore, the fluid liberated into the peritoneal cavity may cause an anaphylactic reaction.

### Summary

The case of a 78-year-old woman with hydatid cysts of the liver is presented. Symptoms were due to pressure of the massively enlarged liver. The complement fixation test, the hemagglutination test and the Casoni test were all positive. Treatment consisted of excision of the smaller pedunculated cyst and marsupialization of the huge cyst after evacuation of approximately 1500 daughter cysts. The cyst wall was treated with Formalin. Despite complications which included temporary biliary fistulae, wound infection, atelectasis and pneumonitis and genitourinary tract infection a favorable outcome was achieved.

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