Multiple Peritoneal Hydatid Disease after Rupture of a Multivesicular Hepatic Hydatid Cyst. Case report

Eugen Tarcoveanu, Gabriel Dimofte, Costel Bradea, Felicia Crumpei, Raluca Anton, Radu Moldovanu

1st Surgical Clinic, “St. Spiridon” University Hospital, Iași

Abstract

We report the peculiar case of a young woman with hepatic hydatid cysts, with numerous peritoneal disseminations (56 cysts) incidentally diagnosed during a caesarian section. The case was managed, surgically preceded and followed by systemic treatment with albendazole. Surgical treatment addressed both the hepatic cyst and the peritoneal hydatid disease aiming to preserve involved abdominal organs.

The diagnosis of peritoneal hydatid disease is today more accurate due to the new imaging techniques and the surgical procedure should be tailored to each patient depending on size, location and complications of each cyst. Radical treatment is the best and represents a goal, but with multiple disease, a staged treatment and special care for organ preservation should prevail as recurrences are not unusual.

Key words
Peritoneal hydatid disease, liver hydatid cyst

Introduction

Hydatid disease is relatively frequent in our country and non-invasive ultrasonic imaging techniques have made possible an earlier diagnosis prior to serious complications. Non-symptomatic hydatid disease may present with complications, but unusual locations as well as multiple primary or secondary hydatid disease pose special therapeutic challenges.

In the 1st Surgical Clinic, in the last 40 years, we operated on 1164 patients with hydatid disease: 764 with hepatic involvement, 240 with pulmonary involvement and 160 with unusual locations. Synchronous multiple cysts represent less than 2% of all cases. Peritoneal hydatid disease is very unusual, especially when it is discovered incidentally. Anatomical and clinical characteristics as well as therapeutic problems raised by this recent case determined us to present it.

Case report

A 26 year young female patient was referred to our surgical department with pain in the right quadrant and lower abdomen, associated with fatigability, lack of appetite, weight loss, constipation, nausea and vomiting. Although symptoms developed slowly in time, her records mentioned a caesarian section three years previously, during which a peritoneal hydatid cyst was found adjacent to the uterus body.

Abdominal examination showed moderate meteorism and low abdominal tenderness. In the epigastrium we identified a round, mobile, elastic tumor 5-6 cm in diameter. There were two similar tumors 10-15 cm in diameter in the right and left lower quadrants. Both were also accessible to vaginal examination, but there was no clear relationship with the uterus. The liver was significantly enlarged having the inferior border 6 cm below the ribs in the mid-clavicular line.

Hematological data showed a slight anemia and a marked increase in the eosinophile count (10%). All other lab exams were within normal limits except for a positive test for antibodies against Echinococcus granulosus.

Liver ultrasound revealed multiple hydatid lesions ranging from 5 to 10 cm in diameter, involving all hepatic segments (Fig.1). Cysts were in different stages of development, uni- or multivesicular, segment VI cyst appeared to be fissured, while the ones developed in segments II and IV had ultrasonic aspect suggestive of infection. The spleen contained a 74/40mm large cyst projected in the hilum and extended towards the inferior pole (Fig.2). Pelvic ultrasound described a cystic structure developed probably
in the mesosigma, 122/123 mm in diameter, and two larger cysts in the right iliac fossa associated with right ureterectasia (Fig.3). A multivesicular cyst was noted in the Douglas pouch (Fig.4), in close proximity with another similar lesion developed on the left lateral aspect of the uterus. Thoracic X-Ray examination excluded a pulmonary hydatid disease and the CT-scan confirmed the presence of numerous cystic lesions 3 to 10 cm in diameter spread in both liver lobes, splenic hilum and the peritoneal cavity. Bilateral ureterectasia was proven but both kidneys appeared to have normal excretion.

We decided for a 10 day preoperative treatment with albendazole in order to insure protective parasiticidal doses in the peritoneal cavity during the surgical procedure.

Surgical exploration confirmed the preoperative diagnosis of the major lesions, but also identified numerous cysts, 27 cysts ranging from 1-12 cm in diameter, developed in the larger omentum (Fig.5). A large number of cysts had developed on the peritoneal surface in almost every anatomical region. Most of them were in-block resected with the large omentum. Cysts developed on the greater gastric curvature posed significant difficulties due to the proximity of the splenic hilum. Ideal total cystectomy could be accomplished with preservation of the spleen. Similarly all 6 cysts involving the Douglas pouch were resected without opening. A 16 cm large multivesicular cyst developed toward the retroperitoneal space and the left broad ligament. We decided for chemical inactivation and subtotal pericystectomy, abandoning a small area of the wall adjacent to the left ureter. The cyst that developed in the mesosigma was chemically inactivated and treated conservatively in order to preserve the sigmoid vessels (Fig.6). Cysts on the lesser omentum had developed between the left lobe and the lesser curvature of the stomach, but also in the bursa omentalis invading the anterior surface of the pancreas (Fig.7) requiring either complete resection or subtotal pericystectomy. We attempted complete resection or chemical inactivation of all visible cysts developed on the remnant peritoneal surface. Altogether 56 hydatid cysts 1 to 6 cm in diameter were neutralized during the procedure (Fig.8).
Hydatid disease of the liver was massive with numerous large cysts, most of them multivesicular, some already infected or associated with major biliary fistulas. Cysts developed in segment IV and in the coronary ligament also invaded the diaphragm. Left hepatic lobe was almost entirely occupied by a 16 cm large cyst with major biliary fistulas. We decided for a conservative approach limiting the amount of hepatic tissue to be sacrificed. All cysts were first chemically inactivated using hypertonic serum, evacuated, followed by a thorough exploration of the cavity (Fig.9).
Fistulas were looked for and sutured and pericyst was partially resected in order to reduce the residual cavity. We considered drainage using silicon tubes in close proximity to the cavities after making them communicate. Gallbladder was removed to facilitate access to a large segment IV multivesicular cyst (Fig.10). We performed an atypical heptatectomy (segment II and III) to deal with the large residual cavity in the left hepatic lobe and the large biliary fistula.

The patient’s recovery was favorable but prolonged, with large amounts of bile exteriorized through the drainages. Antibiotic therapy was directed by bile culture. The patient required an endoscopic sphincterotomy that rapidly decreased the biliary output. She was discharged in the 17th postoperative day while the last drain was withdrawn on an outpatient basis on the 28th postoperative day. A postoperative ultrasound scan showed a 4 cm diameter cyst projected over the right adrenal gland and one 3 cm in diameter in the superior pole of the spleen. Albendazole was continued postoperatively for two more months with periodic liver test evaluation. The 6 month follow-up showed no changes in the splenic and adrenal cysts and a good recovery with excellent general status and weight gain.

**Discussion**

Hydatid disease is caused by Taenia Echinococcus which, in its larval stage, induces the development of a cystic tumor. It is situated most frequently in the liver, followed by the lung and unusual localizations (spleen, peritoneum, kidney, muscle, adrenal gland, ovary, pancreas, thyroid gland, pleura, diaphragm, brain and others) (1). Secondary hydatid disease while unusual (2,3) is generally multiple in location (4).

Symptoms vary according to anatomic location and preoperative diagnosis requires a complex work-out including plain abdominal X-Rays, ultrasound scan, computerized tomography and serological tests.

Peritoneal hydatid disease represents an uncommon occurrence and has been previously described during pregnancy (5). The diagnosis can be suggested by the presence of abdominal cysts in a patient known with liver hydatid disease. Today an accurate evaluation is possible because of the new imaging techniques, which are often able to show specific signs of hydatid disease (6). Even so multiple hydatid disease should be thought of and included in the differential diagnosis of any cystic tumor in solid organs or other anatomic sites, especially in endemic countries (7).

Particular in this case was the absence of symptoms until the caesarian section when an unusual ovarian cyst was discovered. Most probably the primary lesion was a hepatic hydatid cyst fissured in the peritoneal cavity with multiple seeding. This theory is supported by the presence of complicated hepatic lesions, some partially evacuated, with major adhesions around the liver. Lack of any symptoms, including anaphylactic ones, makes it more likely that the initial hepatic cyst produced a small fissure that communicated with the peritoneal cavity with fractionate discharges of hydatid content during a longer period of time.

Therapy raises problems, and though good results have recently been obtained with benzimidazole derivatives, surgical excision is currently the only curative treatment available (8).

Medical treatment is indicated only in cases with multiple cysts inaccessible to surgery, or as a complementary therapy used to reduce the risks of secondary seeding during operation, and prevent recurrences (9, 10). The treatment of choice is surgical and the surgical gesture must be as radical as possible considering the benign character of the disease (11). While complete removal of viable structures is the main goal – with the lowest recurrence rate (12) - its feasibility is mostly related to the location of the cyst (5). In the long term, the high frequency of peritoneal recurrences makes prolonged supervision necessary (13).

We adopted a radical attitude whenever the particular lesions made it possible, but some locations required a more conservative approach. Proximity of the ureter prevented the complete resections of a large mesosigma cyst, while proximity of the pancreas did not pose special problems. The multiple hydatid involvement of the liver was managed in a classic manner, each cyst being treated as an entity. Inactivation of the parasite is indicated prior to opening the cavity after making them communicate. Gallbladder was removed to facilitate access to a large segment IV multivesicular cyst (Fig.10). We performed an atypical heptatectomy (segment II and III) to deal with the large residual cavity in the left hepatic lobe and the large biliary fistula.

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seeding and control the development of residual disease. The accessibility to anti-parasitic medication justifies our conservative attitude regarding involved abdominal organs.

We reported this case because of its peculiarities: a young woman with hepatic hydatid cysts, one of them probably broken in the peritoneal cavity, with numerous secondary lesions, with adequate surgical treatment doubled by pre- and postoperative treatment with Albendazol. Residual disease will be followed by planned ultrasound scans and additional surgery will be scheduled if required.

In conclusion, hydatid disease should be included in the differential diagnosis of cystic masses in solid organs or other anatomic sites, especially in endemic countries. Surgery associated with Albendazol treatment remains the treatment of choice, offering a good clinical result and an acceptable recurrence rate.

References