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Peritoneal Echinococcosis: Anatomoclinical Features and Surgical Treatment

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Abstract
Background Peritoneal hydatid disease is a rare and poorly known disease. We report our experience with 17 patients treated for peritoneal echinococcosis. The main objectives were to analyze and discuss the specific features of the anatomic peritoneal lesions along with their surgical treatment.
Methods We analyzed retrospectively all patients treated in our unit for peritoneal echinococcosis between January 2001 and December 2008. Preoperative description of the lesions, and the surgical procedures were carefully reported.
Results Peritoneal hydatidosis represented 6.3% of all abdominal localizations. There were 10 women (58.8%) and 7 men (41.2%). Median age was 34 years. Ten patients (58.8%) had had previous surgery for abdominal echinococcosis. Ten patients (58.8%) had synchronous abdominal localization of hydatid disease, and two patients had synchronous pulmonary localization. Sixteen patients were operated on by laparotomy. We classified the anatomic lesions into four groups: localized form ($n = 6$, 37.4%), disseminated form ($n = 8$, 50%), "hydatid carcinomatosis" ($n = 3$, 18.7%), hydatidoperitoneum ($n = 1$, 6.25%). One patient had a ruptured hydatid cyst of the left liver. We performed total cystectomies in 10 patients, partial cystectomies in 8 patients, and omentectomy in 5 patients. Two patients (12.5%) had surgical complications. One patient (6.25%) died owing to a pulmonary embolism. Anthelmintic chemotherapy was given to two patients before surgery and to nine patients postoperatively. Recurrences were seen in two patients (14.2%).
Conclusions Peritoneal echinococcosis can cause a large variety of specific and complex anatomic lesions. The disseminated form is the most common, and therefore the surgical treatment is challenging in most cases.

Introduction

Hydatid disease (HD) is a zoonotic infection frequently caused by *Echinococcus granulosus*. Eggs ingested by intermediate hosts, such as humans, liberate embryos in the duodenum, which pass through the intestinal mucosa and enter the portal circulation. Most of these embryos are trapped in the liver, and the rest pass through the liver and reach other organs [1]. The liver and the lungs are the most commonly infected organs (in 60 and 20% of the cases, respectively), but the disease can be seen anywhere in the body [2, 3]. Peritoneal localization represents a rare state [4]. Its features are poorly known because it is mostly reported in the English-language literature as case reports, or it is not distinguished from other extrahepatic abdominal localizations.

In this article, we report our experience with 17 consecutive patients treated for peritoneal hydatid disease. The emphasis is on describing and discussing the specific

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features of multiple anatomic lesions of peritoneal echinococcosis and their surgical management.

Patients and methods

Patients

We analyzed retrospectively all patients treated in our unit for peritoneal echinococcosis between January 2001 and December 2008. Peritoneal echinococcosis was defined as all peritoneal localization of HD not involving solid intraperitoneal organs. Hydatid cysts (HCs) involving the retroperitoneum were excluded. We established preoperative diagnosis by medical history, clinical examination, the usual imaging [ultrasonography (US) and computed tomography (CT)], and hydatid serology. US was routinely used in all patients, and CT was used in case of disseminated disease or difficult surgical access localizations. Intraoperative anatomic lesions and surgical procedures were carefully reported. Topography, size, and macroscopic aspect of all the peritoneal cysts were described and summarized in four main groups.

Surgical procedure

The surgery started with a large incision allowing wide and meticulous exploration of the whole abdominal cavity. The choice of incision depended on the preoperative imaging and was designed in a way that all disease-bearing areas could be easily accessed. Cysts were treated starting with the largest and most complicated ones to reduce the risk of iatrogenic rupture or contamination. The area surrounding cysts was cautiously protected during all the steps of the procedure with packs immersed in 3% hydrogen peroxide. The choice between radical (total cystectomy) and conservative (deroofing) treatment depended on the size and localization of the lesions and was left to the surgeon's intraoperative judgment. When a conservative treatment was chosen, drainage of the residual cavity was mandatory. All cysts were treated in the same setting if possible. Chemotherapy based on albendazole (10 mg/kg per day) was delivered in case of disseminated hydatid disease.

Results

During the period of study, 268 patients were treated in our unit for abdominal echinococcosis, 17 of whom had peritoneal localization (6.3%): 10 women (58.8%) and 7 men (41.2%). The median age was 34 years (range 25–54).

Ten patients had undergone previous surgery for abdominal echinococcosis: eight for liver HC and two for peritoneal HCs. Of these ten patients, two were operated on twice and all others once. The median delay between the first surgery and the diagnosis of recurrence was 72 months (range 24–300 months). All previous surgeries were done in other hospitals and the patients then referred to our unit when recurrence was diagnosed. Of the 17 patients, 1 by mistake underwent a pleural tap for a pulmonary hydatid cyst that was complicated 5 months later by peritoneal echinococcosis.

The most common clinical symptoms (Table 1) were abdominal pain in 13 patients (76.5%) and an abdominal mass in 8 patients (47.1%). Ten patients had synchronous abdominal localization of hydatid disease; it was hepatic in all of them, splenic in three patients, and in the anterior abdominal wall in one. Two patients had synchronous pulmonary HC. The last 5 patients of the 17 had isolated peritoneal echinococcosis (three of them had had previous surgery for hepatic HD and two for peritoneal HD). Hydatid serology was performed in 10 patients and was positive in 8 of them.

One patient did not accept the surgical treatment, and he was treated exclusively with anthelmintic chemotherapy. A total of 16 patients underwent laparotomy (13 by midline incision and 3 by subcostal incision). During surgical exploration, various anatomic lesions were identified, depending on number, macroscopic features, and site. We intentionally grouped them in four main categories (Table 2).

Table 1 Peritoneal echinococcosis: clinical symptoms

Symptoms	No. (%)
Abdominal pain	13 (76.5)
Abdominal mass	8 (47.1)
Fever	4 (23.5)
General symptoms	2 (11.8)
Thoracic pain	2 (11.8)
Jaundice	1 (5.9)
Nephritic colic	1 (5.9)
Vomiting	2 (11.8)
Ascitis	1 (5.9)

Table 2 Peritoneal echinococcosis: intraoperative anatomic lesions

Type	No.	%
Localized form	6	37.4
Disseminated form	8	50.0
Hydatid carcinomatosis	3	18.8
Hydatidoperitoneum	1	6.25

- *Localized form* ($n = 6$; 37.4%): Three patients had one peritoneal HC, and the others had two to six HCs. The cysts were localized around the liver, around the spleen, and in the great omentum. Cyst size varied from 6 to 10 cm.
- *Disseminated form* ($n = 8$; 50%): The peritoneal cavity was filled with multiple cysts occupying all abdominal quadrants. Their size varied from 1 to 15 cm. These cysts had multiple adhesions with the surrounding structures, which made their dissection difficult.
- *Hydatid carcinomatosis* ($n = 3$; 18.8%): This is a definitive form, characterized by millimetric cystic lesions disseminated on all surfaces of the peritoneal serosa (parietal peritoneum, mesentery, mesocolon). It has the same morphology as secondary peritoneal carcinomatosis lesions.
- *Hydatidoperitoneum* ($n = 1$; 6.25%): There is a collection occupying the right part of the peritoneal cavity (Fig. 1), surrounded by an inflammatory membrane, isolating it from the surrounding structures. This collection was connected to a right hepatic HC.

In one patient, surgical exploration revealed a ruptured HC of the left liver lobe, with free daughter cysts in the peritoneal cavity. Cyst rupture occurred several weeks before surgery and was asymptomatic. Left lobectomy with ablation of all the daughter cysts and extensive peritoneal lavage were performed.

The perihepatic sites were most frequent (Fig. 2), and were found in eight patients (Table 3). In theses cases, the cysts were subhepatic and had strong adhesions to the liver and diaphragm. Five patients reported a history of previous hepatic HC surgery, one patient had synchronous HC of the liver, and one had a history of basal right pleural tap.

We performed total cystectomy in 10 patients, partial cystectomy or deroofing in 8 patients, and omentectomy in 5 patients. These procedures were often combined in the same patient. In the case of hydatidoperitoneum, complete dissection of the membrane from the surrounding structures was difficult, provoking several injuries of the small bowel. It was the reason surgical abstention was finally decided. Associated procedures are summarized in Table 4. The median operating time was 160 min (60–345 min). The two patients with synchronous lung HC underwent thoracic surgery 3 months before abdominal surgery.

The median hospital stay was 12 days (4–16 days). Surgical complications occurred in two patients (12.5%). The first patient was reoperated on postoperative day (POD) 1 for hemorrhagic shock due to a bleeding from a short gastric vessel, which was easily ligated. The second patient was reoperated on for a pelvic collection. One patient (6.25%) died on POD 1 due to acute respiratory distress following pulmonary embolism.

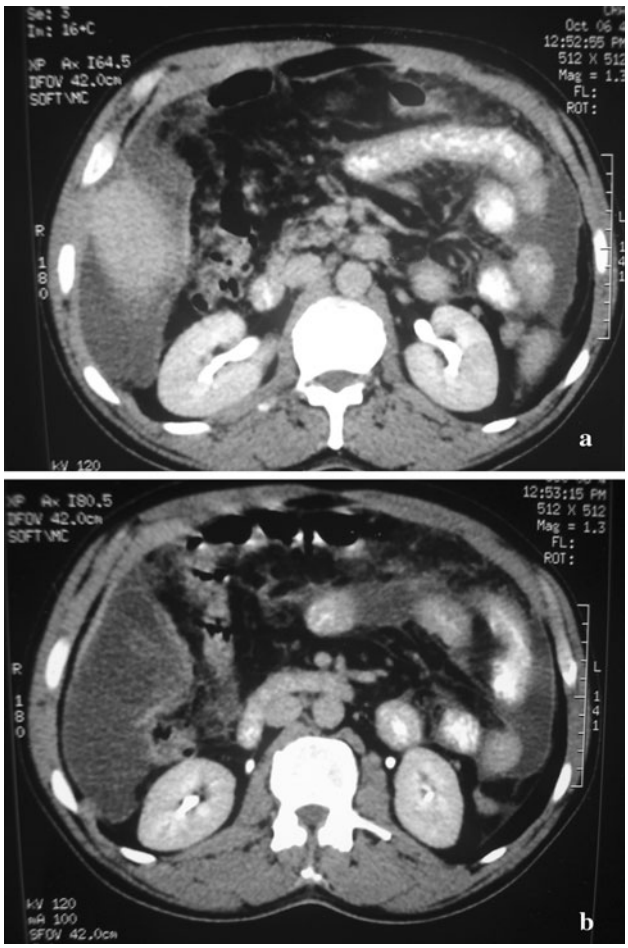

Fig. 1 a, b Computed tomography shows radiologic aspects of hydatidoperitoneum, a right peritoneal collection connected to a right liver hydatid cyst

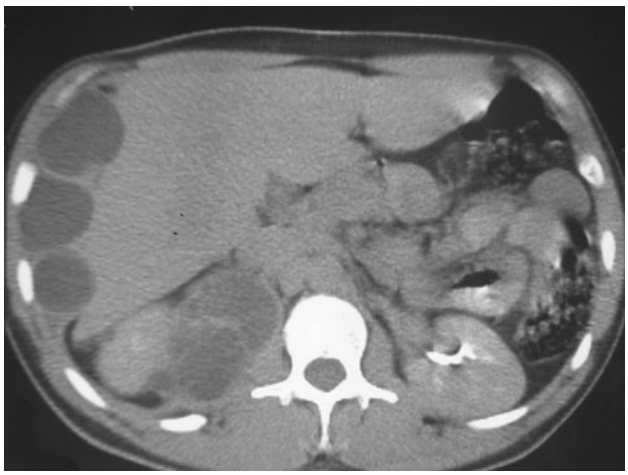

Fig. 2 Computed tomography shows perihepatic peritoneal HCs

Anthelmintic chemotherapy (albendazole 10 mg/kg per day) was given to two patients for 3 months before surgery and to nine patients for 6 months postoperatively.

Table 3 Peritoneal echinococcosis: hydatid cyst localization

Localization	No.
Perihepatic region	8
Right paracolic region	2
Pelvis	6
Left paracolic region	1
Left subphrenic region	2
Splenic hilum	1
Great omentum	5
Mesentery	2
Mesocolon	3
Hydatid carcinomatosis	4

Table 4 Peritoneal echinococcosis: associated surgical procedures

Procedure	No.
Left hepatic lobectomy	2
Unroofing of liver HC	6
Splenectomy	1
Unroofing splenic HC	2
Cystectomy abdominal wall	1

HC hydatid cyst

After a median follow-up of 37 months (12–96 months), recurrences occurred in two patients (14.2%). The first patient developed peritoneal and pulmonary HC 5 years after the initial surgery. He was first operated on by the thoracic approach; 3 months later he was then managed for his abdominal cyst. Medical treatment was given 3 months before and 6 months after surgery. After 1 year, CT showed a retroperitoneal HC. The second patient developed an asymptomatic peritoneal recurrence 6 months after surgery. He had hydatid carcinomatosis during the first surgery and was managed by medical therapy. The two others patients with hydatid carcinomatosis had normal abdominal US at 12 and 13, respectively.

Discussion

The peritoneum remains an uncommon site of HD, even in endemic countries. The frequency of this localization varies from 1.4 to 9.4% of all localizations [5]. In our series, the peritoneum was involved in 6.3% of abdominal localizations, which is similar to other Moroccan reported studies [4, 6]. In the literature, intraperitoneal HC represents up to 13% of abdominal HDs [2].

Peritoneal echinococcosis is secondary to hepatic infection in 66 to 75% of cases [7]. The pathophysiology of secondary peritoneal echinococcosis is unclear. This dissemination could be explained by microrupture, which is mostly asymptomatic, or by leakage of echinococcal fluid during surgery, which causes peritoneal contamination in 5–14% of cases [4, 8]. Small quantities of hydatid fluid are discharged in the large peritoneal cavity. Vesicles and scolices may become encysted, producing a vesicular encysted peritoneal hydatidosis, or remain free, causing real hydatid carcinomatosis [4]. In all of our patients, peritoneal hydatidosis was secondary and hepatic infection was the main cause (76.4%). Ten of our patients (58.8%) had undergone previous surgery for HC and were all operated on in other hospitals before being referred to us after the diagnosis of recurrence. This suggests that a lack of surgical expertise could be a risk factor for secondary peritoneal HC following surgical intervention. Primary peritoneal echinococcosis is rare, with few reported cases [1, 8–11]. Peritoneal HC is considered a primary localization when there are no HCs at other sites [12]. It may be caused by hematogenous spread through the arterial circulation [13].

Surgery is the only curative treatment for peritoneal HD [4, 10]. According to the guidelines of the World Health Organization (WHO), peritoneal localization is an indication for surgical treatment [10]. Small peritoneal cysts that are asymptomatic may be managed conservatively. Surgical intervention is required for symptomatic and large peritoneal cysts [10].

Subserous implantation with development of peritoneal HCs is the most common form of peritoneal echinococcosis [4]. It represented 87.4% in our study and 76.0% in the study by Benamer et al. [4]. In all, 50% of our patients had the disseminated form. The cysts occupy the entire peritoneal cavity and sometimes numbered more than 100 [4]. The localized form is characterized by the presence of one or a limited number of HCs. It represented 37.4% in our study and 15.0% in the study by Benamer et al. [4]. Perihepatic localization was most frequent (50%) and followed primary hepatic HD in most cases. The surgical procedures should be customized to each patient depending on the size, location, and complications associated with each cyst [14]. It must be performed safely to avoid intraoperative morbidity and mortality [15]. The surgeon should treat complicated and large cysts first [4]. Total cystectomy, whenever possible without organ sacrifice is the treatment of choice [16]. Drainage and wide deroofing is recommended when cysts are attached to intraperitoneal viscera or major vessels, or in case of deep localization [5, 8]. Removal of all the cysts during the same intervention is preferred [17]. Otherwise, planned reintervention should be considered [15].

In three patients (18, 7%), we found millimetric HC disseminated in all the surface of the peritoneum. We call it

"hydatid carcinomatosis" because it resembles disseminated peritoneal carcinomatosis. Benamer et al. found this form in 6% of his patients [4]. The diagnosis is always made peroperatively. Because of this widespread dissemination, no surgical treatment can be proposed in such cases [4, 18].

One patient had a hydatidoperitoneum. It is a rare and particular form that is found in up to 18% of peritoneal HD [4]. More often secondary to a ruptured hepatic HC, it is characterized by creation of a new membrane that surrounds the hydatid material and the intraperitoneal viscera, isolating them from the rest of the abdominal cavity [4]. During the early stage of an inflammatory granuloma, the membrane is highly inflammatory and is adherent to the surrounding structures. Any attempt to remove it is extremely dangerous (which was the case in our patient). In this situation, it is recommended that we start a medical therapy and wait and few months until the stage of "adventitial maturation" is reached, when the membrane becomes fibrotic and thus more easily and safely removed. In such cases, treatment of the primary cyst is mandatory during the same surgical intervention [19].

Intraperitoneal rupture is a rare complication of hepatic HD. In the literature, the incidence varies from 1.75 to 8.60% [20–22]. The rupture can be spontaneous, but it is more frequently secondary to trauma [20]. In one patient, there was an asymptomatic rupture of a hepatic HC, discovered operatively several weeks later. Intraperitoneal rupture is usually symptomatic, with constant abdominal symptoms and inconstant allergic ones [21]. The occurrence of sepsis, acute abdomen, intraperitoneal bile leakage, or shock calls for emergent surgical management, including treatment of the primary cyst, evacuation of all daughter cysts, and thorough peritoneal lavage. In the absence of complications, surgery may be postponed until a more appropriate time [23].

According to the WHO guidelines, chemotherapy is recommended for peritoneal cysts. The reduction of clinical signs together with the changes in cyst appearance and a reduced number of viable cysts following chemotherapy [24, 25] suggest its usefulness in the management of patients with single or multiple HCs located in one or more organs. Preoperative use of albendazole or mebendazole can reduce the risk of cystic echinococcosis recurrence [24]. Moreover, it softens and reduces intracystic pressure, thereby simplifying cyst removal [26]. Polat et al. [27] reported that both preoperative and postoperative use of albendazole might help to prevent recurrence. In our practice, we use medical therapy in cases of disseminated hydatid disease. Until recently, there has been no formal consensus concerning the modality and the duration of the treatment [28], and there is a need to establish comprehensive guidelines for medical treatment of peritoneal hydatidosis [3].

As most cases of peritoneal hydatidosis are secondary to intraoperative spillage, precautions should be used to minimize its occurrence [29]. A rigorous surgical technique is recommended, including wide protection of surrounding organs, the use of a scolicidal agent, and careful aspiration of hydatid cyst fluid in cases of deroofing. The benefit of using medical therapy in the case of intraoperative spillage to prevent peritoneal recurrence is still unclear. Many authors recommend the use of anthelmintic treatment to reduce the risk of recurrence when there is cyst spillage during surgery, partial cyst removal, or biliary rupture [3]. The WHO recommends postoperative chemotherapy in cases in which spillage may have occurred: albendazole for at least 1 month or mebendazole for 3 months [30].

To our knowledge, at the time of preparing this manuscript our study represents the first series in English-language publications that reports and describes all possible anatomoclinical forms of peritoneal hydatidosis. We have proposed the surgical management for each form based on our own experience and a review of the literature.

Conclusions

Although rare, our study shows that peritoneal echinococcosis can cause a large variety of specific and complex anatomic lesions. Complete resection of all of the lesions in the same setting is the best option for curative treatment but is not always possible. The surgeon must adapt the procedures according to the type and localization of the lesions and the patient.

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