

Case Report

Diagnosis and Surgical Approach in 76 Cases of Hepatic Hydatid Cysts in Adults and Children

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Keywords

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- Diagnosis
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Abstract

Background: The high prevalence of hepatic hydatid cyst (HHC) still remains one of the major public health problems in our country.

Patients and Method: In the last 30 years, 76 patients with HHC have been operated on in our department. Their clinical and pathological features, diagnosis, surgical treatment and outcome are retrospectively analyzed.

Results: The study group comprised 47 (61.8%) males and 29 (38.1%) females. The age of the patients varies between 8-72 (range 44) years. Pediatric cases presented clinical and therapeutical peculiarities. Diagnosis was established by physical examination, serology, ultrasonography and rarely CT scan. Fifty-five cysts were located in the right lobe, 15 cysts in the left lobe and 6 cysts were bilateral. Twelve patients had multiple cysts and 3 patients had co-existing cysts with different abdominal or lung locations. There were 6 cases with recurrent disease. Majority of our cases had complications i.e. infection (n=15), rupture in the biliary duct without or with obstructive jaundice (n=13), rupture in the peritoneal cavity with acute abdomen (n=2), another co-morbidity (n=5). All patients underwent surgical treatment but in 6 cases the laparoscopic approach has been successfully used. Surgical procedures were partial cyst pericystectomies with external (n=60) or internal (n=2) drainage in 62 cases, total cyst pericystectomies in 7 cases, and left hepatic (atypical or segmentary) resections in 7 cases. The common bile duct was explored in 11 patients and it was drained by a T-tube in 6 cases or by choledocoduodenostomy in three others. Papilosphincterotomy was practiced in two patients. Chemotherapy (albendazole) was sporadically used after surgery. We had no deaths but a significant rate of morbidity (n=15; 19.7%) with a prolonged hospital stay.

Conclusion: Surgery remains the mainstay treatment of HHC. The topography and volume together with the great number of complicated cases and co-existing pathology forced us to a variety of surgical approaches most of them conservative. Our results were encouraging with acceptable morbidity and no lethal cases for the study period.

BACKGROUND

Cystic echinococcosis (hydatid disease) is a parasitic infestation by a tapeworm of the genus *Echinococcus* (E) type. Two of the recognized species *E. granulosus* and *E. locularis* are important for humans. The disease occurs worldwide being endemic in the Mediterranean basin and other sheep-raising areas including Romania, as well as large regions of Asia, Australia and New Zealand, Africa and South America [1-3].

Echinococcosis can involve any organ but usually affects the liver (70% of cases), lung (20%) and also the spleen, kidney, the bone or the brain [4]. Beside the high prevalence and incidence HHC raises difficulties of early diagnosis, has unpredictable

clinical course linked to numerous complications and disposes of more therapeutic alternatives, most of them surgical but are not always optimal for a given case [5,6]. Diagnosis of HHC is based on epidemiological and professional inquiry, physical examination, serum serologic testing for antibodies against hydatid antigens but mainly on imaging with abdominal ultrasound or CT/MRI [7,8]. Reported also mainly in endemic area HHC in children evolves asymptomatic or with nonspecific features for long time, diagnosis being fortuitously, within a complication or due to anaphylactic phenomena. The goal of management of HHC is to neutralize and withdraw the parasite with minimal morbidity and mortality. Surgery is the golden standard of therapy describing several operative approaches although there is not

an optimal conservative or radical technique used in current practice. Conservative procedures (cyst removal and partial cyst pericystectomy) aren't usually yet used by many practitioners being somewhat easier to perform but associated with raised morbidity, recurrence rate and prolonged hospital stay [9,10].

Radical procedures including total cyst pericystectomy and hepatic resections achieved spectacular successes, canceled the risk of recurrences but had higher intraoperative risks [11]. There are also two treatment alternatives: chemotherapy with low cure and high recurrence rates but useful as associated method and interventional procedure – PAIR (puncture, aspiration, injection and reaspiration). They are preferred in cases found in children in the last years laparoscopic surgery has been successfully proposed at any age but in selected cases [12-14].

PATIENTS AND METHODS

Between 1987 and 2016, 76 patients with HCC were diagnosed and operated on in our surgical tertiary unit. There were 47 men (61.8%) and 29 women (38.1%) ranged in age from 8 to 72 years with an average age of 44 years. Seven cases were under 16 years. Five patients had previous surgery for HCC (all of them operated on elsewhere) before being referred to our unit. Clinical, laboratory and imaging studies, types of surgery and operative protocols, results and outcome were retrospectively analyzed.

RESULTS

Clinical presentation is shown in Table (1). Abdominal pain and tenderness were the most common complaints followed by a palpable mass usually in the right upper quadrant. Intermittent fever and jaundice were possible proving they had biliary communications with their cyst resulting in their extrahepatic biliary obstruction due to daughter cysts or infection. Additional features were pruritus, rash or urticaria or anaphylactic phenomena. Two cases presented signs of acute abdomen while five "asymptomatic" cases were accidentally discovered on a routine ultrasound or intraoperatively during cholecystectomy for lithiasis.

Fifty-five cysts were located in the right lobe, 15 cysts in the left lobe and 6 cysts were bilateral. The diameter of the cysts were between 5-17 (range 8) cm. Twelve patients had multiple cysts (between 2 and 5). The results of current liver tests were modified in 40 cases (52.6%) eosinophilia and elevation of alkaline phosphatase being the most common abnormalities. Classical Cassoni's test was positive in 25 initial cases. Later indirect hemagglutination, immunoelectrophoresis and especially enzyme-linked immunosorbent assay (ELISA) for IgG against Ag % or Ag B furnished the best confirmative results for biological diagnosis. Imaging. Chest and abdominal X-ray gave diagnostic images in few typical circumstances showing orientating evocative calcifications or hepatomegaly. Due to its wide availability, low cost and high diagnostic rate ultrasound is the first diagnostic attempt (Figure 1).

According to the Gharbi's ultrasound modified classification of HHC types, describes proper the different lesional aspects (Table 2). Computerized tomography was utilized in only few cases having previous surgery or disease complications

Presenting clinical features	No of patients
Abdominal pain/tenderness	34 (44, 7%)
Hepatomegaly, abdominal mass	23 (36, 2%)
Dyspepsia	16 (21,5%)
Anaphylactic event: pruritus, rash, urticaria	10 (13, 1%)
Intermittent fever	9 (11, 8%)
Intermittent jaundice	8 (10, 5%)
Acute abdomen (cyst rupture, liver abscess)	2 (2, 6%)
"Asymptomatic" incidental discovery	5 (0, 65%)

No %: Co-morbidity included co-existent lung (n=2) or splenic (n=1) hydatid cyst (resolved in one- or two-stage operations) (15), pulmonary embolism (n=1), pleural effusion (n=2), cholelithiasis (n=7), diabetes mellitus (n=5), HTA (n=8), ischemic heart disease (n=6), overweight and/or obesity (n=5) [15].



Figure 1 Nine cm Ø CHH in the right lobe (segm 5-6).

Type 1 (unilocular cystic lesion, uniform anechoic content, cyst wall visible)	14 (18,4%)
Type 2 (multivesicular, multiseptated cysts, sand & debris)	23 (30,2%)
Type 3 (cyst containing liquid with floating membrane, septa x/& daughter cysts)	22 (28,9%)
Type 4 (cyst with heterogenous hypo/hyperchoic content)	12 (15,7%)
Type 5 (cyst with partial thick calcified wall)	5 (6,4%)

identifying internal septa, daughter cysts within the main cyst and intra-cyst debris or calcifications. It had a high sensitivity being an important tool to determine vascular, biliary or extrahepatic extension and recognize complications such as infection and rupture and therefore to assess respectability (Figure 2). All 76 patients from our study underwent surgical treatment based on the topography, size and the presence or absence of complications or co-morbidity. In the majority group including 62 patients we performed conservative procedures consisting in partial cyst pericystectomy (Mabitt-Lagrot) removing the

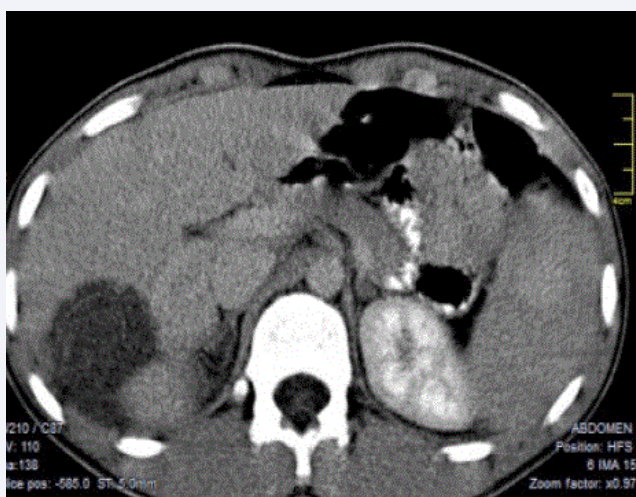


Figure 2 CT of the right lobe sided CHH.

anterior wall of the cystic lesion as widely as possible together with all of its contents assuring also external drainage. The narrowing of the residual cavity was realized by capitonnage or introflexion method suturing the outer surface layers of the cyst to each other or to the bottom of the cavity keeping the cyst walls folded. Internal efficacious drainage of residual cavity was assured by Roux en Y-loop pericystojejunostomy. In two large, centrally located cases with important bilio-cystic fistula for which fistula closure or hepatic resection could not be performed. Intraoperatively after cyst evacuation we noted presence of a biliary fistula in 11 cases and cyst rupture in the bile duct in 4 cases associated with jaundice and dilatation or debris in major bile ducts. Identification and removal of parasitic remnants with intraoperative radiological or instrumental (ERCP) exploration was mandatory and T-tube insertion in the main bile duct (n=6), bilioenteric shunt (n=2) or papilosphincterotomy (n=2) was always necessary.

In fewer circumstances we appealed to radical surgical techniques undergoing three enucleore sections (Imperati) and four total "ideal cyst pericystectomy (Napalkov) in small size lesions (< 5-6 cm), located peripherally and mostly exteriorized. Finally in 7 "happy opportunities" of single, convenable sized cysts taking the biggest hepatic lobe space a left lobectomy or bisegmentectomy were performed all of them with a simple evolution. In one case of "spontaneous" ruptured HCC our 34-year-old male patient without any history of systemic disease or trauma an emergency laparotomy for abdominal pain, nausea, vomiting associated with guarding and rebound tenderness was required. Baseline investigations were normal except leukocytosis. Ultrasound showed a 7 cm Ø lesion with irregular borders and some cystic areas in the left lobe of the liver. Minimal fluid was detected in perihepatic areas and lower abdomen. During surgery a 7-8 cm cyst with a tear on the inferior surface was identified. Seropurulent fluid and some daughter vesicles were also founded through abdominal cavity. The patient underwent a partial cystectomy and the peritoneal cavity was washed out with hypertonic saline and drained. Postoperative course was uneventful and he commenced abendazole, therapy which continued 4 months after his discharge. No recurrence

or additional pathology was detected at the 6- and 12-month follow-up. In the last decade 6 patients underwent laparoscopic management of HHC after careful selection of unique small- sized (5-6 cm), uncomplicated, superficially located (left lobe or V-VI segments) cysts. Method was effective and reliable. Cystotomy and partial cystectomy with drainage were successfully practiced in convenient operative time (range 80 min) without morbidity or conversion. Laparoscopic surgery appeared much less traumatic to the patient and with a better cosmetic effect (Figure 3). Our experience provided satisfactory clinical efficacy and good incidence of cure, acceptable rates of complications, no (known) own recurrences and a reasonable mean length of hospital stay.

Postoperative complications were dominated by biliary and infectious events. The most of these unfor-tunate events appeared after traditionally verified procedures but only a postoperative cavity abscess appeared after laparoscopic surgery (P<0,045). Four biliary leakages were conservatively treated in three cases draining < 100 ml/24h and by endoscopic sphincterotomy in one low-put fistula. Also four cases needed re-operation because of hemorrhage and infection in cystic cavity. Residual cavity abscess manifested in by fever and purulent drainage in 5 cases was solved by antibiotics, minimal surgical drainage and lavage. Our morbidity rate was acceptable due to reduced incidence of abdominal wound but also to general complications. The average hospital stay was 15 (range 10-65) days. The mean follow-up period was 24 (range 12-36) months and no recurrence was reported in our series during this period.

Statistical analysis

In order to assess rates and means of differences within each group of patients and cysts we have applied the Chi square test

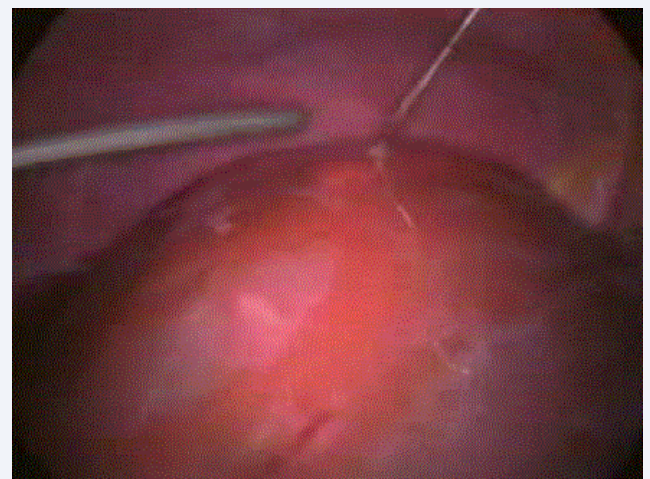


Figure 3 Intraoperative aspect of laparoscopic approach of HHC.

Table 3: Postoperative complications.

Biliary leakage	4 cases
Infection of cavity (abscess)	5 cases
bile ascites	1 case
Pleural effusion	4 cases
pulmonary emboli	1 case

and X2 test. Running the analysis we conclude that P value of less than 0.05 was considered to indicate statistical significance.

DISCUSSION

HHC lesional complexity continues to pose countless problems of management especially in the surgical treatment related to operative technique and tactics. Modern therapeutic concepts of the disease are oriented to knowledge and understanding the hepato-biliary pathophysiology surgery aiming not only at healing patients but also at cancelling as many anatomoclinical sequels as possible. Early diagnosis of this zoonosis is determinative taking into account the evolution and prognosis of the condition but also predominantly young, active socio-professional patients are affected [16,17]. Although our experience is not wide it illustrates that in time the evolution of the diagnosis and treatment of HHC. In the past diagnosis of HHC was based on history and clinical findings, x-ray imaging and sometimes on serendipity, discovering sometimes earlier uncomplicated cases with important details for the surgical act but also in most instances significantly evolved lesions [18]. Actually epidemiological, clinical, biological and especially imaging data, allowed early diagnosis of this disorder however in our series as in other statistics coming from endemic regions the percentage of advanced and complicated cases still remains important [8,19].

Thereby out of our many patients only 5 of them were completely asymptomatic and 26 cases had more or less troubling manifestations but the rest of 45 sufferings bearing a HHC presented annoying, dominant, painful even and acute signs and symptoms. Defining for most experiences in HHC treatment is that surgery abides the mainstay method, medical therapy alone and PAIR remains only complementary approach in particular circumstances [20].

For the majority of practitioners surgical management procedures have continuously evolved over years but together with considerable controversies about the most effective technique and also about the current place of surgery among other therapeutic options [21]. The first dilemma concerns the use of conservative, classical procedures performed in majority of our own cases - 81,5% - against radical techniques that we used in only 18.5% of cases.

Comparative retrospective study between these two surgical practices described significantly higher morbidity and recurrence rate in the first group ($P < 0,045$). We registered 9 specific complications e.g., 4 infections of the residual cavity and 5 biliary fistulisation, events having similar proportions of other authors.

Partial cyst pericystectomy proved a safe procedure in most of our cases with more than satisfactory immediate and in time results, non prohibitive morbidity and zero mortality rates. Control and drainage of main biliary duct was necessary in 11 cases, through a T-tube drainage, choledocoduodenostomy or ERCP. In terms of rigorous selection radical surgery was less often performed total cyst pericystectomy and also left lobar or bisegmental exeresis in 7 cases each was practiced when possible due to the unique, small size ($< 5\text{cm } \emptyset$) uncomplicated lesion with superficial exteriorized topography. However radical

surgery provides an effective surgical management option in preventing local recurrences and cavity related complications but must be carried only in tertiary units by experienced hepatobiliary surgeons [22,23]. Because of limited experience in many centers, controversies about the laparoscopic treatment of HHC sometimes still persist [24,25]. However in strict selected uncomplicated cases as in the 6 personal observations method proves feasible and safe.

Finally the surgeons who practice this pathology should expect an extreme variation of lesions and should know and should be able to execute the whole range of surgical procedures. Our study recognizes some limitation due to retrospective character and small number of patients with different severity of the disease operated on in an extended period of time. Thus we cannot compare results of different methods of surgical treatment and the place of medical therapy is unconvincing. In addition another possible limitation is an inadequate patchy long-term follow up of operated cases.

CONCLUSIONS

Hepatic hydatid disease remains a common medical and social problem in our country and especially in our regional area. Our experience covered all lesional register of this pathology and in time we practiced the majority of consecrated surgical procedures and in the last years even laparoscopic surgery has been successfully tried. Despite anatomical and clinical diversity of our series summing simple but also complicated cases, postsurgical results were satisfactory not recording a prohibitive morbidity and zero mortality rate.

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