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Surgery for hepatic hydatidosis
Endoscopic treatment of complications

Cystic hepatic hydatidosis, caused by the tape-worm *Echinococcus granulosus*, is an ancient disease, endemic in sheep rearing areas. Despite improvement in conservative treatment, it remains a surgically treated disease for the good risk patient. It may present complicated and challenging problems in liver surgery and there continues to be controversy regarding the appropriate surgical technique. A series is presented of 200 patients, suffering from cystic hepatic hydatidosis, operated on consecutively from 1985 to 2001, applying the same operative procedures consisting of wide capsectomy and drainage. This surgical technique is described in detail. There was a mortality rate of 0.5% and postoperative complications occurred in 14%, mostly related to bile leaks. Endoscopy was selectively performed for biliary fistulas, taking into account the volume and duration of biliary drainage. They all healed and no re-operation was required. There was recurrence of echinococcus in 11 of the 115 patients monitored for from 4 to 20 years (i.e. 10%). Drainage operations, when properly performed, present excellent results regarding total extirpation of hepatic hydatidosis. Few postoperative complications and a low rate of late recurrences are observed, and the need for more aggressive surgical approaches is, in this way, reduced. Endoscopy may be successfully applied for treating long-lasting biliary fistulas following surgery.

C. Vagenas,
C. Spyropoulos,
C. Stratis,
J. Maroulis,
C.E. Vagianos
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Department of Surgery, Rion University Hospital, Patras, Greece

Χειρουργική θεραπεία της ηπατικής εχινοκοκκίασης. Ενδοσκοπική αντιμετώπιση των επιπλοκών

Περίληψη στο τέλος του άρθρου

Hydatidosis or echinococciasis is a parasitic disease known since antiquity, when "livers full of water" were mentioned by Hippocrates ("hydor"/ύδωρ, meaning water in Greek). The disease is scattered throughout the world, its cystic form being endemic in areas where sheep are raised, including the Mediterranean basin, Australia, South America, the Far East, Middle East and Eastern Europe.[1] Despite a significant reduction of hepatic hydatidosis in Greece, due to an official campaign against this zoonosis over the past 20 years,[2] the disease is still present, in the form of both recurrences and new complicated cases, and may pose challenging surgical problems.

The infection is caused mainly by the tape-worm *Echinococcus granulosus* and less commonly by *Echinococcus multilocularis*. The latter is confined to the northern hemisphere, represents less than 5% of total echinococcal disease and is practically never seen in the Mediterranean area. It is characterized by a different infection pattern,[3] and presents a more virulent and aggressive form of echinococciasis, resulting in liver invasion by multiple cysts, and is much more difficult to cure.[4]

Echinococcal disease is cyclozoonotic and the parasite has a biphasic life cycle, first described in 1862 by Leuckhard and Heubner,[5] requiring two hosts or carriers to complete its development. The cysts in humans are mainly located in the liver (70–75%), almost three quarter of them in the right lobe, and the lungs (20–25%), while rarely, location in almost any solid organ and even bones has been reported.[6]

The diagnosis of hepatic hydatidosis is today almost exclusively based on imaging techniques such as ultrasonography (US) and computed tomography (CT). When interpreted by experienced radiologists these provide significant information regarding the nature of a cystic hepatic lesion, its exact location, its relation to other organs and structures and the morphological characteristics.[7] In doubtful cases, where the differential diagnosis from simple liver cysts, polycystic disease or cystic neoplasms remains difficult, immunodiagnostic tests, such as Casoni's intradermal, indirect hemagglutination, ELISA or complement fixation tests may be of assistance, despite their high percentage of false negative and false positive results, ranging overall from 10% to 40%.[8,9]

At this time, surgery remains the treatment of choice for hepatic hydatidosis in the good risk patient. The aim is to remove the parasite totally and treat the remaining cavity, avoiding bile leaks, which are responsible for most of the immediate postoperative complications. Prevention of the consequences of disease recurrence, either locally in the liver or in the peritoneum, by applying meticulous surgical technique and administering complementary medical treatment, are among the prerequisites of surgery for hepatic hydatidosis. The aim of this paper is to present a type of "aggressive" drainage operation for the surgical treatment of hepatic hydatidosis, which has been successfully applied at Rion University Hospital, Patras for the last 20 years.

MATERIAL AND METHOD

Between January 1985 and December 2001, 200 consecutive patients suffering from hepatic hydatidosis were treated surgically at the Department of Surgery of Rion University Hospital, in Patras. Details about the patients and the characteristics of the cysts are presented in table 1. Twenty-four of these patients (12%) had previously undergone one or more operations for hepatic disease. In all patients the same type of "aggressive" drainage operation described below was applied.

The commonest presenting symptom was upper abdominal pain (82%), followed by fever. Twenty-four patients (12%) gave a history of jaundice, 9 (4.5%) of allergic reactions, while in 23 (11.5%) the disease was discovered incidentally, being asymptomatic. The diagnosis in all patients was based almost exclusively on CT and US imaging (fig. 1). The Casoni skin test and antiechinococcal antibodies, measured by an enzyme-linked immunosorbent assay, were applied in doubtful cases (25 and 30 cases respectively).

Table 1. Characteristics of patients and cysts.

Characteristics of patients	n=200
Men/women	83/117
Age (years)	16–85 (median 61)
Previous surgery	24 (12%)
Cyst characteristics	
Cyst diameter (cm)	5–25 (median 10)
Monolocular/multilocular	22/178
Lobe: Right/left/bilateral	147/30/23
Infected cysts	72 (36%)

Table 2. Patients operated on for hepatic hydatidosis at the University Surgical Department of Patras, and their postoperative hospitalization at different time periods.

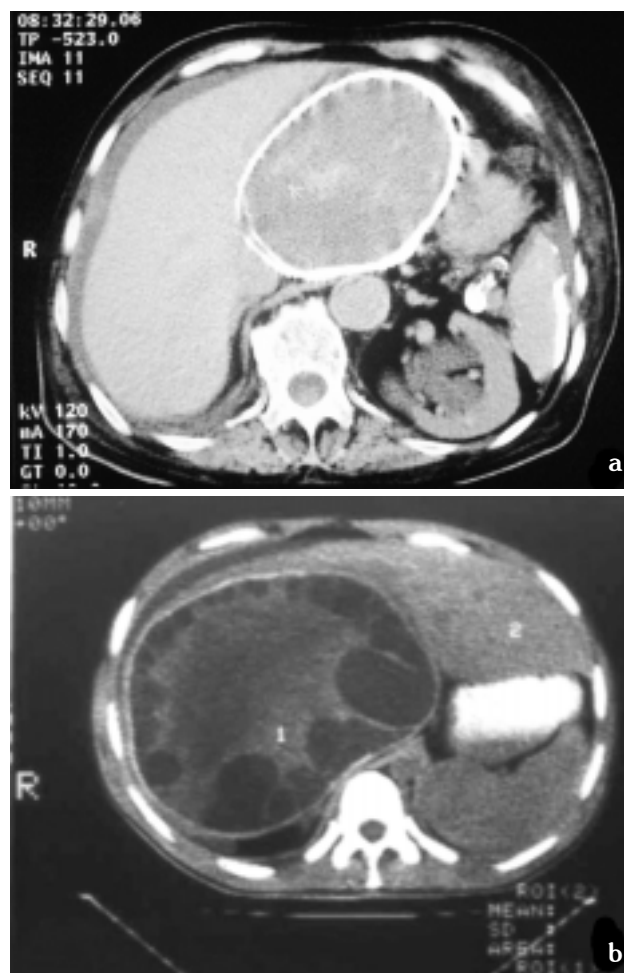| Years	Number of patients	Hospitalization days (mean±SD)
1985–1991	67	11.8±9.2
1991–1995	81	14.8±12.3
1996–2001	52	11.4±7.2
	Total: 200	

Figure 1. Typical computed tomography findings in hepatic hydatidosis. (a) Monolocular cyst with calcified pericyst. (b) Multilocular hydatid cyst.

Surgical technique

An extended right subcostal incision is employed, including upper vertical and left subcostal extension when technical difficulties are encountered. The liver is fully mobilized by dividing the hepatic ligaments and the cyst

is totally exposed. The incision and the peritoneal cavity are carefully isolated with compresses soaked in scolicocidal hypertonic saline solution (15% NaCl) in order to avoid peritoneal contamination (fig. 2). During the whole procedure special care is taken to avoid peritoneal spillage of hydatid fluid, which may result in an immediate anaphylactic reaction and later peritoneal recurrence. The cyst is punctured initially with a special Y-shaped tru-cut and hydatid fluid is removed in order to reduce intracystic pressure (fig. 3). An opening on the surface of the cyst is made; daughter cysts and further hydatid fluid are removed. Finally the cyst is completely deroofed by performing the widest possible capsectomy and the cavity edges are oversewn with an interlocking braided suture of either polyglycolic acid (Dexon) or polyglactin 910 (Vicryl), in order to avoid bleeding and bile leaks. In the event of cholelithiasis when the gallbladder is located near the cyst or when there are cystobiliary communications, a cholecystectomy is performed. A cholangiogram through the cystic duct will show the presence of duct pathology (small daughter cysts, hydatid debris and, uncommonly, stones). In such cases the bile duct is explored. Cystobiliary communications are identified by infusing normal saline through the cystic duct after performing the Pringle maneuver and obstructing the peripheral common bile duct with an atraumatic clamp. Backflow of the saline will reveal any communications, which are then sutured with 3/0 Dexon or Vicryl (fig. 4). The inner surface of the residual cavity is carefully searched by inspection and US for identification of small satellite cysts, which are simply drained through the cavity wall (fig. 5). In the case of complicated, multilocular cysts, the possibility that satellite cysts are present in the liver parenchyma is extremely high, so intraoperative US, either trans-hepatic or through the remaining cavity, is used to reveal their presence and location. Omentoplasty is not performed as a routine, but only according to the surgeon's preference.
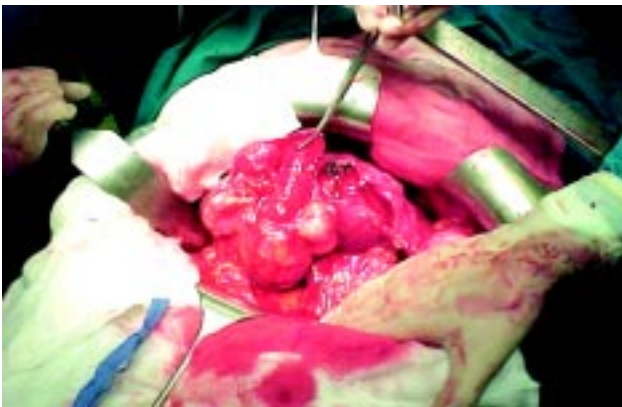

Figure 2. Isolation of the surgical field with compresses soaked in hypertonic saline.

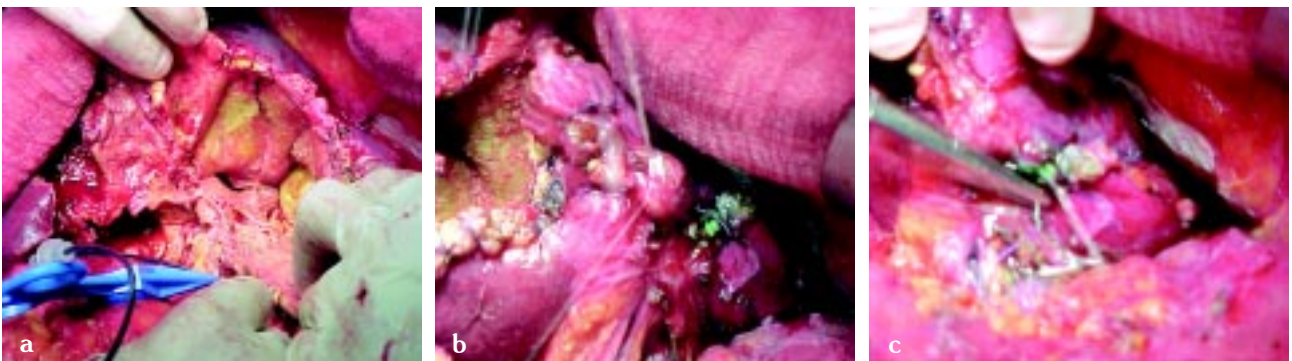**Figure 3.** Evacuation of the cyst content results in reduction of intracystic pressure.

Figure 4. Identification and treatment of cystobiliary communication. (a) A catheter is inserted into the cystic duct and after performing Pringle maneuver, normal saline is injected with pressure. (b) The cystobiliary communication is easily identified in the residual cavity by the backflow of the injected saline. (c) The communication is obliterated by suturing it with 3/0 absorbable suture.
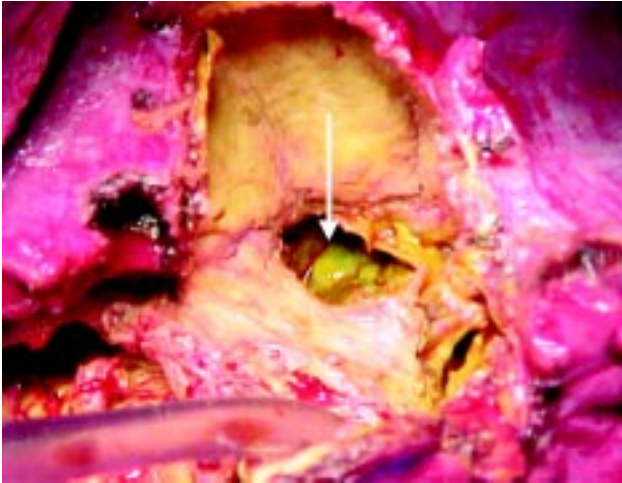
Figure 5. Drained satellite cyst. The cyst (arrow) after being identified by intraoperative ultrasound, is drained through the pericyst of the remaining cavity.

The cavity is drained by one or two negative-pressure, closed system drainage systems (Jackson-Pratt bulb drain, Cardinal Health Medical Products, Ohio, USA), often temporarily anchored at the desired position in the remaining cavity by 3/0 Plain Cat Gut suture.

Adjunct treatment. Anthelminthic drugs (imidazole compounds) are administered two weeks preoperatively, in an effort to sterilize the cyst fluid and for three months postoperatively, even if there has been no obvious intra-operative spillage of hydatid fluid. At the beginning of the series mebendazole was administered (50 mg/kg of body weight) while now albendazole is given exclusively (10 mg/kg of body weight).

The diagnosis of postoperative bile fistula was based on clinical examination, laboratory findings and imaging techniques. After confirmation of a fistula and verification of complete external drainage of bile, based mainly on US, the patients were subjected to conservative and supportive treatment, depending largely on the amount of bile excreted and the duration of the fistula. As a routine, high output fistulas (>300 mL bile/day) of more than one week's duration without signs of reduction were treated endoscopically, as were low output fistulas (<300 mL bile/day) of more than three weeks' duration without signs of reduction. An endoscopic retrograde cholangiopancreatography (ERCP) was performed in order to reveal and treat the cause of the increased intrabiliary pressure, but even if no specific cause was found, endoscopic sphincterotomy, with or without insertion of a stent or a nasobiliary catheter under low suction (10–15 cmH$_2$O), immediately reduced the

amount of bile excreted and helped the fistula to heal in a very short time.

Follow-up of the patients was based on US and CT six months after the operation and every two years thereafter in the asymptomatic patient. Recurrence was defined as hydatid disease discovered in the liver on US and verified by CT. CT was also necessary for the verification of peritoneal recurrence.

RESULTS

A significant reduction in the number of patients with hepatic hydatidosis treated in the Department has been noted since 1985. The postoperative hospital stay was shortened over this time, mainly due to the increased surgical experience and the optimized treatment of complications (tab. 2). Various diagnostic imaging techniques and blood tests were evaluated. It was clearly shown that CT and US were very effective in giving the correct diagnosis (sensitivity 96% and 93%, respectively), while the Casoni skin test and antiechinococcal antibodies, applied only in doubtful cases, were found positive in 60% and 70%, respectively of proved cases of hydatidosis. Eosinophilia was present in 15% and increased bilirubin in 16%, and almost 36% of the patients had laboratory and clinical evidence of suppuration of the cyst contents.

Cholecystectomy was performed in 79 patients (40% of the total). Bile duct disease was found in 24 cases (12%), in all of which the common bile duct was explored, cleared of small daughter cysts, hydatid debris or biliary stones and drained by a T-tube. Omentoplasty was arbitrarily performed in 41 cases (20% of the total).

The mortality and morbidity rates are shown in table 3. Only one patient (0.5%) died, an 81 year-old man who died on the fourth postoperative day due to a cerebrovascular accident. Twenty-seven of the patients (14%) suffered from immediate postoperative complications, 20 of which (10% of the total) were related exclusively to bile leaks. Three patients developed subdiaphragmatic abscesses, due to malfunction of the suction drain, 2 of which were drained percutaneously and one surgically. Two developed biliary peritonitis on the first postoperative day and had to be reoperated on. In one this was due to an accessary bile duct in the gallbladder bed and in the other to ineffective drainage of a cystobiliary communication. Fifteen patients developed biliary fistulas, one of them bronchobiliary. Of these fistulas, nine (60%) healed by expectant management, most

Table 3. Postoperative complications in 200 patients.

Complications	Number of patients	Percentage (%)
Bile leak-related	20	10
Biloma/abscess	3	1.5
Bile peritonitis	2	1
Bronchobiliary fistula	1	0.5
Biliodermal fistula	14	7
Intraoperative anaphylactic shock	2	1
Pneumonia	3	1.5
Postoperative hemorrhage	1	0.5
Myocardial infarct	1	0.5
Death	1	0.5

Table 4. The effect of omentoplasty on postoperative hospitalization time and incidence of biliary complications (NS: not significant).

	No omentoplasty (159 patients)	Omentoplasty (41 patients)
Hospitalization days		
Mean±SD	11.6±9.6	12.8±7.8
Median (range)	9 (3–94)	11 (4–40), NS
Biliary complications	17	3, NS
Fistula	14	1, NS
Biloma/abscess	2	1, NS
Bile peritonitis	1	1, NS

of them as outpatients. They required an average of 30 days until the drain could be removed. The remaining six (40% of all fistulas), including the bronchobiliary fistula, were subjected to ERCP as previously described. They all healed within two weeks and the drain was removed shortly after (fig. 6). This meant that 18% of the low and 100% of the high output fistulas required some kind of endoscopic intervention.

Table 4 shows that there was no significant benefit regarding either hospital stay or postoperative morbidity by the addition of an omentoplasty to the procedure.

A complete follow-up, ranging from 4 to 20 years was accomplished in 115 patients (58% of the total). There were recurrences in 11 patients (10% of those subjected to adequate follow-up), 7 of which were located in the liver and four peritoneal.
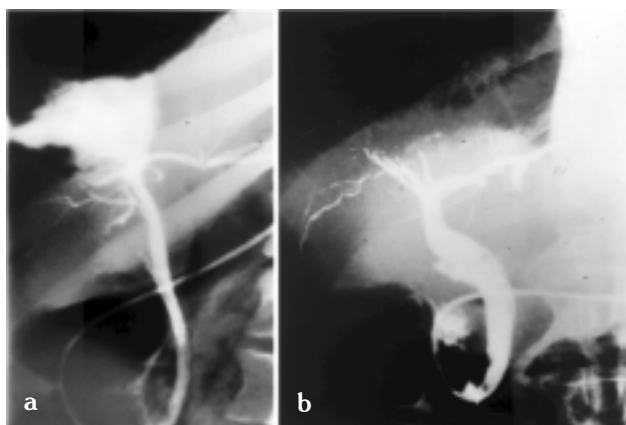

Figure 6. Treatment of a biliary fistula by placing a nasobiliary catheter. (a) Before treatment. Obviously there is massive leak of bile into the remaining cavity. (b) Fifteen days after treatment, the leak has completely stopped.

4. DISCUSSION

Hepatic hydatidosis has a worldwide distribution, being more common in rural sheep-rearing areas with suboptimal hygienic conditions, where close proximity of living between its intermediate (man, sheep, cattle, etc.) and definitive carriers (mainly dog) helps the maintenance of the parasite's life cycle. At present surgery remains the preferred treatment for the good risk patient, and although reports have been published of successful laparoscopic approaches in selected cases of hepatic hydatidosis,[10,11] open laparotomy is still the approach of choice. Alternative, less invasive techniques, such as percutaneous drainage of the cysts followed by injection of sclerosing agents or hypertonic saline, have also been reported recently with good results.[12,13] However, the dire consequences of peritoneal dissemination, namely anaphylaxis and peritoneal recurrence, as well as the frequent presence of multi-septated cysts containing thick fluid, resulting in ineffective evacuation of the cyst contents, render surgeons skeptical and unwilling to apply this new treatment approach widely.

Numerous surgical approaches have been applied for the effective extirpation of the parasite, and there is still controversy regarding the best.[14] They can be divided into "radical" and "conservative" surgical procedures. Radical operations attempt to remove the cyst totally, en bloc with the pericyst, which is the outer adventitial layer, formed by the compressed and fibrotic liver. They include pericystectomies and hepatectomies.[15] Conservative operations attempt to neutralize the parasite and evacuate the contents of the cyst without removal of the pericyst.[16]

Despite unnecessary loss of hepatic parenchyma, it has been claimed that radical operations in experienced hands reduce bile leaks, shorten hospital stay and re-

duce recurence, mainly by removing the so called "exogenous" or satellite daughter cysts, responsible for local recurrence.[17]

It is the authors' opinion that radical surgery for the treatment of hepatic hydatid cysts constitutes over-treatment for a benign liver disease. A significant amount of hepatic parenchyma is removed unnecessarily, particularly in hepatic resections, while pericystectomy for centrally located cysts, adhering to major vessels is far from being considered as a safe surgical procedure.

The alternative method of performing partial pericystectomy has the advantage of avoiding intraperitoneal spillage, by removing an intact cyst.[15] Resection can be reserved for penduculated or small peripheral cysts. "Conservative" drainage surgery, when properly performed, is considered as the method of choice for treating large hepatic hydatid cysts.[18]

The role of omentoplasty in the surgical treatment of hepatic hydatidosis remains controversial.[19,20] In this series, no significant advantage regarding either hospital stay or postoperative morbidity, was gained by adding omentoplasty to the standard procedure. Consequently this technique is nowadays considered optional for surgery of hepatic hydatidosis at this Department.

The major complications of surgery include hemorrhage and the leak of bile from the communications between the cavity and the biliary tree. Cystobiliary communications are quite common, while the incidence of intrabiliary rupture of cysts has been reported to reach 25%.[7,21] After drainage, a pressure gradient facilitates bile flow through these communications towards the cavity rather than into the duodenum.[22] This bile leak represents the main source of immediate postoperative complications. If not properly drained, it may result in the formation of abscesses in the residual cavity or passage to the peritoneum and subsequent bile peritonitis. If drained effectively outwards, an external biliary fistula will develop and this is the commonest complication.

It seems that by using closed system negative pressure drains, better results are achieved in terms of draining all fluid outwards and avoiding external contamination of the residual cavity, a common and greatly feared complication in the past.[18] There are hardly any indications for open surgery nowadays for the treatment of external biliary fistulas. Endoscopic intervention with sphincterotomy, biliary stenting, dilatation of strictures or nasobiliary drainage in the case of persistent fistulas has eliminated the need for the complex reoperations of

the past.[23] Even when formation of bronchobiliary fistulas takes place, a complication extremely difficult to manage and associated with high rates of morbidity and mortality, the insertion of an endoprothesis with a following reduction of the intrabiliary pressure has shown excellent results, reducing the need for surgical intervention.[24]

Recurrence of the disease, either in the peritoneal cavity or locally in the liver, represents a significant complication of surgery for hepatic hydatidosis, often requiring hazardous reoperation.[25,26] This is caused either by intraoperative spillage of hydatid fluid into the peritoneal cavity or by incomplete removal of the cyst or cysts. Almost 30% of patients with operative dissemination are expected to develop secondary peritoneal echinococcosis,[27] while between 5% and 10% of all operated patients are expected to develop post-operative recurrence.[28] The problem of satellite cysts has been addressed by Magistrelli et al,[18] and their incidence has been reported to reach 29.5%.[29] They are vesiculations of daughter cysts beyond the pericyst layer, not identifiable at the operation. These cysts as well as small "occult" liver cysts, not detectable during the procedure, appear to be responsible for local, hepatic recurrence. Low recurrence rates have been reported after pericystectomy in several series[30,31] and many authors consider drainage operations to be associated with higher recurrence.[16,18] However, rates of less than 6% have been reported with carefully performed drainage operations.[17] Careful preoperative evaluation of the CT-scans by experienced radiologists provides the exact location of all the primary cysts, including the satellite cysts. Operative US also can be helpful in detecting these small satellite cysts, particularly when performed through the remaining cavity.[32] The authors consider operative US absolutely necessary for cysts highly suspicious of harboring satellite cysts, such as multilocular, infected or multiple liver cysts.

Initial conservative treatment of asymptomatic recurrent small cysts seems to be effective,[33] but symptomatic or exceptionally large peritoneal cysts should be treated surgically before complications develop, and surgery in such cases may need to be repeated several times until permanent eradication is achieved.[25] Total cystectomy, whenever possible, represents the treatment of choice for peritoneal cysts, except when they are firmly attached to intraperitoneal viscera, when drainage and wide deroofing is safer and equally effective.[34]

Radical treatment of disseminated peritoneal echinococciasis is not possible. In such cases, only the large,

symptomatic cysts should be surgically removed, anthelminthics should be administered continuously and the patient should be monitored closely. This therapy seems to be effective in keeping the disease asymptomatic for prolonged periods.[25,33]

Cystic hepatic hydatidosis is a worldwide disease, encountered mostly in areas with sub-optimally equipped hospitals and doctors often not experienced in major hepatic surgery. It is therefore of great importance to propose a safe procedure such as that described, offering results equally as good as major resection, and associated with significantly less mortality and morbidity. The application of endoscopic techniques for the prevention or treatment of unavoidable complications offers additional safety and eliminates the need for the major and complex operations of the past, even in more specialized centers, with significant experience in extensive liver surgery.[35]

ΠΕΡΙΛΗΨΗ

Χειρουργική θεραπεία της ηπατικής εχινοκοκκίασης. Ενδοσκοπική αντιμετώπιση των επιπλοκών

К. ΒΑΓΕΝΑΣ, Χ. ΣΠΥΡΟΠΟΥΛΟΣ, Χ. ΣΤΡΑΤΗΣ, Ι. ΜΑΡΟΥΛΗΣ, Κ. ΒΑΓΙΑΝΟΣ

Χειρουργική Κλινική, Πανεπιστημιακό Γενικό Νοσοκομείο Πατρών, Ρίο, Πάτρα

Αρχεία Ελληνικής Ιατρικής 2006, 23(4):351–358

Η ηπατική εχινοκοκκίαση προκαλείται από το παράσιτο *Echinococcus granulosus* και αποτελεί μια αρχαία νόσο ενδημούσα σε κτηνοτροφικές περιοχές. Παρά τη βελτίωση των συντηρητικών μέσων αντιμετώπισης, η πάθηση εξακολουθεί να είναι κατά κύριο λόγο χειρουργική, συνοδευόμενη συχνά από επιπλοκές, γεγονός που καθιστά ιδιαίτερα σημαντική την ορθή αντιμετώπισή της. Εντούτοις, διεθνώς εξακολουθεί να υπάρχει διχογνωμία σχετικά με την ιδανικότερη χειρουργική τεχνική. Παρουσιάζονται αναδρομικά 200 ασθενείς με εχινόκοκκο ήπατος, οι οποίοι χειρουργήθηκαν στη Χειρουργική Κλινική του Πανεπιστημιακού Νοσοκομείου Πατρών κατά τη χρονική περίοδο 1985–2001. Σε όλους τους ασθενείς εφαρμόστηκε η ίδια χειρουργική τεχνική εκτεταμένης κυψεκτομής και παροχέτευσης, η οποία και περιγράφεται στο κείμενο αναλυτικά. Το ποσοστό της θνητότητας ήταν 0,5%, ενώ μετεγχειρτικές επιπλοκές εμφανίστηκαν στο 14% των ασθενών, κυρίως λόγω χολόρροιας. Η παρουσία χοληφόρων συριγγίων αντιμετωπίστηκε ενδοσκοπικά, με βάση την παροχή και τη διάρκεια του συριγγίου. Ίαση επήλθε σε όλες τις περιπτώσεις, χωρίς να απαιτηθεί επανεγχείρηση. Υποτροπή της νόσου σημειώθηκε στο 10% από τους 115 ασθενείς που υποβλήθηκαν σε ικανοποιητική μετεγχειρτική παρακολούθηση (από 4–20 έτη). Η ορθή εφαρμογή παροχετευτικών επεμβάσεων στην ηπατική εχινοκοκκίαση συνεπάγεται εξαιρετικά αποτελέσματα όσον αφορά στην εκρίζωση του παρασίτου. Το ποσοστό των μετεγχειρτικών επιπλοκών και των υποτροπών είναι μικρό, μη δικαιολογώντας πλέον ριζικές χειρουργικές παρεμβάσεις. Η εφαρμογή ενδοσκοπικών τεχνικών βοηθά στην επιτυχή αντιμετώπιση των χοληφόρων συριγγίων υψηλής διάρκειας και παροχής, που αποτελούν την κύρια μετεγχειρτική επιπλοκή της επέμβασης.

Λέξεις ευρετηρίου: Ενδοσκόπηση, Εχινοκοκκίαση ήπατος, Παροχετευτικές επεμβάσεις, Χοληφόρα συρίγγια

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Corresponding author:

C. Vagianos, Department of Surgery, Rion University Hospital, GR-265 04 Patras, Greece
e-mail: vagian@otenet.gr