

Management of complex liver cystic hydatidosis: challenging benign diseases for the hepatic surgeon

A case series report from an endemic area

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Abstract

Hydatid cysts of the liver are benign lesions which require a wide range of surgical strategies for their treatment. We hypothesized that cysts larger than 15 cm, or compressing main vascular structures, or located in both hemilivers should be considered, as well as complicated cysts, in the category of complex hydatid cysts.

In a retrospective study including 55 patients, we evaluated the characteristics of complex hydatid cysts, and compared surgical outcomes between patients operated on for complex cysts (Complex Group) and those operated on for non-complex cysts (non-Complex Group).

In the Complex Group, 19% of patients had cysto-biliary communication with recurrent cholangitis, 9.5% had cysts eroding the diaphragm or chest wall, or communicating with the bronchial tree, 31% had cysts with contact with main vascular structures, 11.9% had multiple bilobar cysts, 14.3% had giant cysts with organ displacement, and 14.3% had a combination of the above-mentioned types. Type of surgical treatment was different between the two groups ($P < .001$). Additional procedures were statistically more frequent in the Complex Group ($P = .02$). Postoperative morbidity was higher in the Complex Group, although not in a significant manner ($P = .07$). Median hospital stay was longer in the Complex Group (12 vs 7 days, $P < .001$). No 30-day mortality occurred. Four patients (7.3%), all belonging to the Complex Group, required reoperation for postoperative complications.

Surgery for complex hydatid cysts of the liver is potentially burdened by serious complications. This kind of benign liver disease requires skill-demanding procedures and should be treated in centers with expertise in both hepato-biliary surgery and hydatid disease management.

Abbreviations: CT = computed tomography, ERCP = endoscopic retrograde cholangiopancreatography, US = ultrasound.

Keywords: complex cyst, echinococcosis, hydatidosis, liver, surgical treatment

1. Introduction

Hydatid disease, also named as echinococcosis, is a worldwide diffuse parasitic disease, that is endemic in the Middle East, Far

East, South America, Australia, New Zealand, East Africa, and Mediterranean basin countries.^[1–3] *Echinococcus granulosus* is the most common among the *Echinococcus* spp. which affects humans, and is the causative agent of cystic echinococcosis. The parasite has the dog as definitive host. Humans are occasional intermediate hosts who contract the disease by accidentally swallowing the tapeworm eggs, usually contained in contaminated food or water. Development of cysts mostly occurs in the liver, where the protoscolices migrate through the portal flow.^[1,2,4]

Liver cystic hydatidosis usually remains asymptomatic for a long time, until the cyst size enlarges or complications occur. The most common complications are infection and subsequent abscess formation, rupture of the cyst into the biliary or bronchial system, and rupture into the peritoneal cavity.^[5–7] In general, patients with complicated hepatic cysts face serious and sometimes life-threatening clinical situations.^[8] In certain occasions, hydatid cysts remain clinically silent or cause few and non-specific symptoms in spite of the fact that they require prompt surgical treatment because of the risk of sudden and severe complications. The latter type of cysts can also be considered as complex hydatid cysts, due to the need for skill-demanding surgery for their cure, although they have little clinically relevant effects.

The surgical department of our academic institution has been considered the reference center for hepatobiliary surgery and for surgical treatment of hydatid cysts in Northern Sardinia, where

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hydatidosis is endemic.^[9] In the latest decades, we have observed a progressive trend in reduction of surgical interventions for hepatic hydatidosis, mainly due to both reduction of echinococcosis infections and success of non-operative management in most patients. As a result, we observed that surgical treatment of cystic liver hydatidosis of the liver is nowadays almost exclusively reserved to patients with complex cases.

The main purposes of this study were to describe the management of complex hepatic hydatid cysts in an institution located in an endemic area, and also to propose a definition of complex hydatid cyst of the liver.

2. Materials and methods

Data of every patient consecutively submitted to liver surgery has been prospectively collected in an institutional-approved database. The institutional review board of the Department of Medical, Surgical and Experimental Sciences of the University of Sassari approved this retrospective study. For the present study, we queried our database for any patient operated on for hepatic hydatid disease between January 2010 and March 2020. Those with alveolar hydatid cysts were excluded. Before operation, all patients underwent routine blood tests, Boyden test, chest X-ray, liver ultrasound (US), and triphasic liver computed tomography (CT) scan. In selected cases, also cholangio-MRI and/or endoscopic retrograde cholangiopancreatography (ERCP) were carried out. Patients' demographics and cyst characteristics were collected (age, sex, liver location, number, and size of the cysts, US and CT scan characteristics), as well as type of surgical operation, complications, and hospital stay.

For the purposes of this study, we divided the patients into two groups, the "Complex Group" and the "non-Complex Group". We considered in the former patients having at least one of the following conditions:

- cysto-biliary communication with recurrent cholangitis;
- erosion of the diaphragm or chest wall, or communication of the cyst with the bronchial tree;
- contact with or compression of main vascular structures, such as vena cava, hepatic veins, portal vein, or main portal branches;
- multiple bilobar cysts;
- giant cysts with organ displacement.

Type of surgical operation and surgical outcomes were compared between the Complex Group and the non-Complex Group. The patients' demographic and cysts characteristics were summarized with descriptive statistics. Continuous variables were analyzed using the *t* test or Mann-Whitney *U* test, as appropriate. Differences in proportions between the groups were evaluated using the χ^2 or the Fisher exact test, where appropriate. All *P* values lower than .05 were considered to indicate statistical significance.

3. Results

During the considered time frame, 55 patients underwent surgery for treatment of hydatid cyst disease of the liver. Of those, 24 (43.6%) were males, and 31 (56.4%) females. Mean age was 60.6 years (range 19–79). Ten patients (18.2%) presented with recurrent hydatid cysts. Signs and symptoms of the study population are resumed in Table 1. As for the characteristics of the cysts, 18 patients (32.7%) had cysts located in both hemilivers, while 22 (40.0%) had more than one cyst. Mean

Table 1

Characteristics of the patients included in the study.

	N (55)	%
Age (mean, range)	60.6 (19–79)	
Male sex	24	43.6
Symptoms/signs		
Abdominal pain	19	34.5
Palpable abdominal mass	31	56.4
Fever	8	14.5
Jaundice	7	12.7
Nausea/vomiting	6	10.9
Anorexia	20	36.4
Upper abdominal tenderness	5	9.1
Abdominal distension	22	40.0
Recurrent cholangitis	6	10.9
Elevated ALT/AST	15	27.3
Patients with recurrent cysts	10	18.2

cyst size was 10.0 cm (range 3.4–24.0 cm), and 30 (54.5%) patients had at least one cyst measuring 10 cm or more in diameter. Nine patients (16.4%) presented with additional hydatid cyst location other than liver (Table 2). Characteristics of the complex cases, according to our inclusion criteria, were resumed in Table 3. In particular, 8 (19.0%) patients had type I (cysto-biliary communication with recurrent cholangitis) (Fig. 1), 4 (9.5%) had type II (erosion of the diaphragm or chest wall, or communication with the bronchial tree) (Figs. 2 and 3 and Supplementary Material: Video 1, <http://links.lww.com/MD/F294>), 13 (31%) had type III (contact or compression of vascular structures: vena cava, hepatic veins, portal vein and/or main portal branches) (Fig. 4), 5 (11.9%) had type IV (multiple bilobar cysts) (Fig. 5), and 6 (14.3%) had type V (giant cysts with organ displacement) (Fig. 6 and Supplementary Material: Video 2, <http://links.lww.com/MD/F295>). Six patients (14.3%) had a combination of the above mentioned types.

The surgical procedures carried out were: radical pericystectomy (4.8% vs 69.2%), subtotal pericystectomy (14.3% vs 0%), partial pericystectomy (23.8% vs 7.7%), hepatic segmentectomy (45.2% vs 15.4%), right hepatectomy (7.1% vs 0%), and left hepatectomy (4.8 vs 7.7%) in the Complex Group and non-

Table 2

Characteristics of the cysts.

	N	%
Cyst location		
Right lobe	30	54.5
Left lobe	7	12.8
Bilobar	18	32.7
Cyst number		
Patients with single cyst	33	60.0
Patients with multiple cyst	22	40.0
Cyst size (cm) (mean, range)	10.0 (3.4–24.0)	
<10 cm	25	45.5
≥10 cm	30*	54.5
Additional hydatid cyst location other than liver	9	16.3
Lung	6	10.9
Kidney	1	1.8
Pelvis	1	1.8
Peritoneum	1	1.8

* In 6 cases cyst diameter was ≥15 cm.

Table 3**Classification of complex hydatid cyst (42 patients).**

Complex hydatid cyst type	No. of patients	%
I Cysto-biliary communication with recurrent cholangitis	8	19.0
II Erosion of the diaphragm or chest wall, or communication with the bronchial tree	4	9.5
III Contact or compression of vascular structures: vena cava, hepatic veins, portal vein, and/or main portal branches	13	31
IV Multiple bilobar cysts	5	11.9
V Giant cysts with organ displacement	6	14.3
Type I + III	3	7.1
Type I + III + IV	2	4.8
Type I + III + V	1	2.4

Complex Group, respectively. In general, the type of surgical procedure was statistically different between the two groups of the study ($P < .001$). Additional procedures during primary surgery for treatment of liver cystic hydatidosis were statistically

more frequent in the Complex Group (97.6% vs 61.5%, $P = .02$) (Table 4). All those additional procedures became necessary for the complete treatment of the hydatid disease, except for one case in which a concomitant adrenalectomy was planned for a 5-cm right incidentaloma.

Median hospital stay was significantly longer in the Complex Group (12 vs 7 days, $P < .001$). In total, 15 patients (27.3%) developed one or more postoperative complications. Postoperative morbidity was higher in the Complex Group (33.3% vs 7.7%), although not in a significant manner ($P = .07$). Reoperation for occurrence of post-operative complications was needed in 4 patients of the Complex Group. In the non-Complex group, one patient developed a pleural effusion and one an intra-abdominal abscess. The main part of the complications, as expected, was registered in the Complex Group: 1 patient developed pneumonia, 1 an isolated pleural effusion, 1 a wound infection and a pleural effusion, 3 an intra-abdominal abscess and a pleural effusion, 3 a post-operative bleeding requiring re-laparotomy, 1 a colonic ischemia, and 5 an isolated biliary fistula (Table 5). Five patients developed a postoperative biliary fistula, which resolved spontaneously in 2 cases within an average of 7 days. In 2 patients, the biliary fistula was resolved with ERCP and

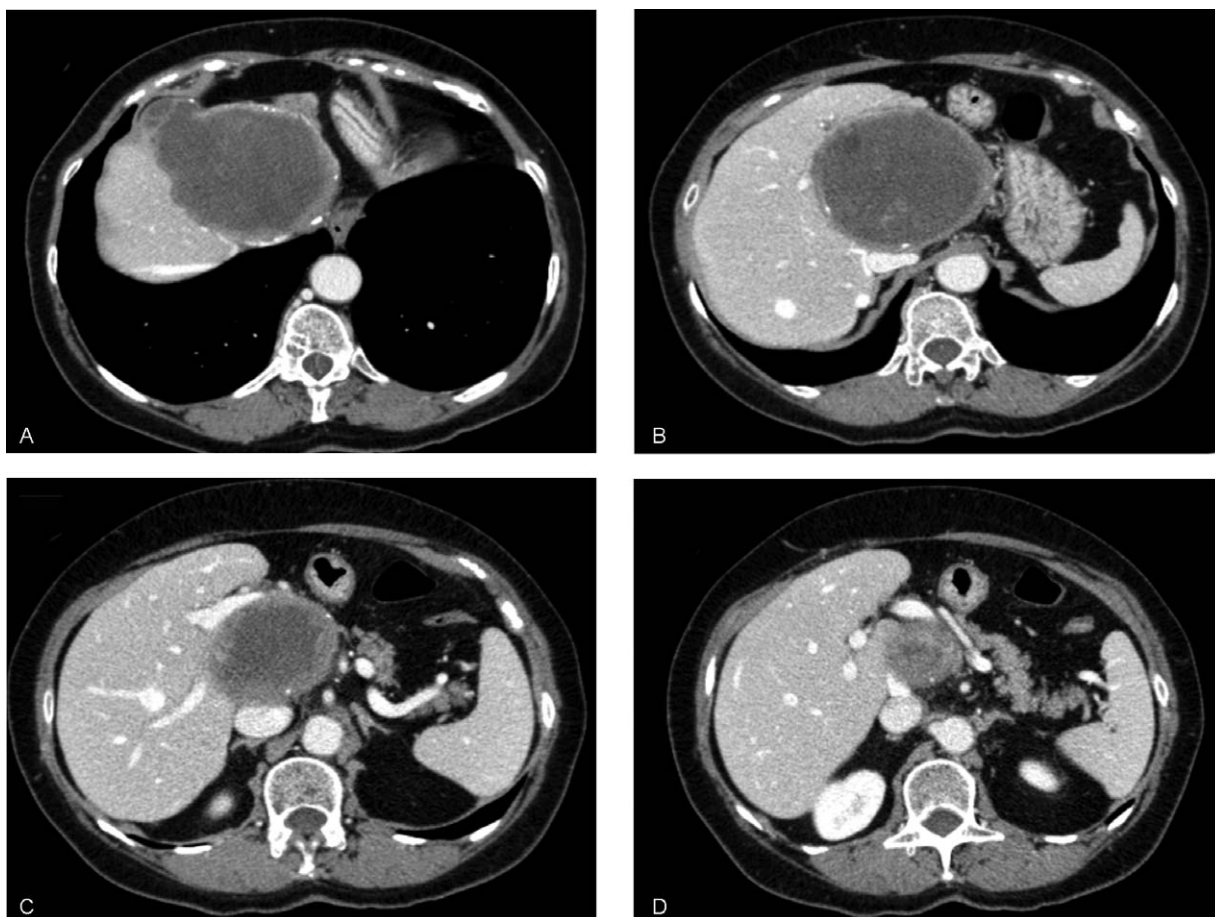


Figure 1. CT scan showing a complex hydatid cyst of the liver in 60-year old woman, presenting with symptoms of recurrent cholangitis due to cysto-biliary communication (A). The cyst caused compression of the inferior vena cava, left and middle hepatic veins (B and C). The inferior aspect of the cyst was in contact with the hepatic hilum, resulting in compression of the portal vein (D). To note, the cyst also caused atrophy of the left lobe. The patient underwent a sub-total pericystectomy, followed by hilar biliary duct-jejunostomy for recurrent episodes of cholangitis after 6 months. After 1 year from the first surgery the patient deceased for sepsis caused by carbapenem-resistant *K. pneumoniae*.

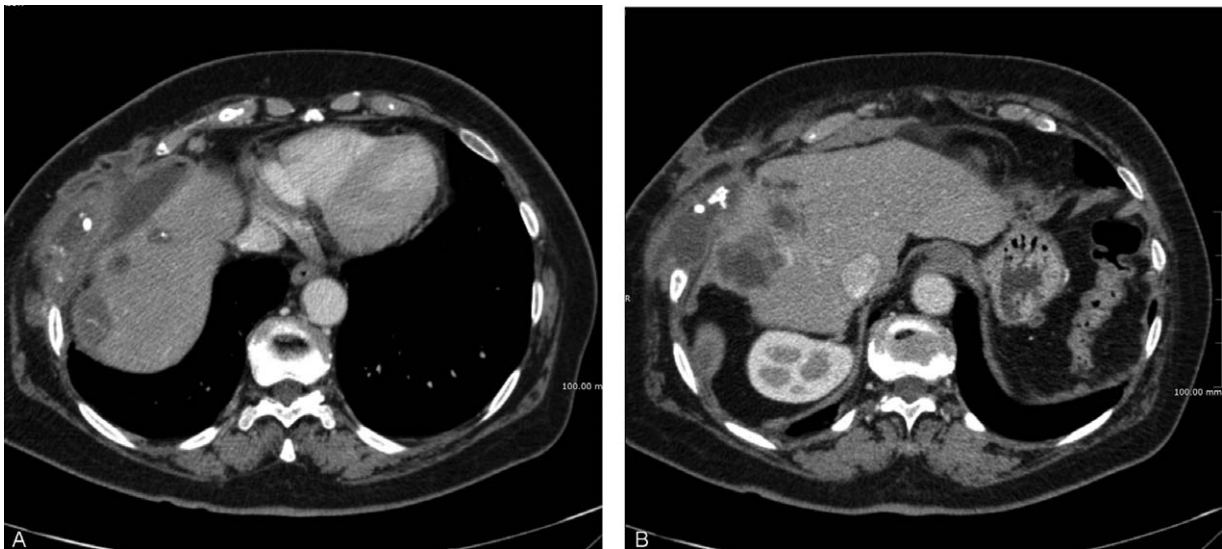


Figure 2. CT scan revealing multiple hydatid cysts of the right hepatic lobe involving the diaphragm, the lower pulmonary right lobe, and eroding the chest wall in a 78-year-old woman (A and B). She underwent a right hepatectomy associated to diaphragm repair with mesh, costal resection, and atypical pulmonary resection.

sphincterectomy, while 1 patient who initially had a biliary-enteric anastomosis necessitated a re-laparotomy for anastomotic fistula repair and drainage.

There was no 30-day mortality. One patient, who initially underwent a partial pericystectomy for a cyst located in close vicinity of the hepatic hilum (Fig. 1), was re-operated after 3 months for persistent episodes of cholangitis and deceased

12 months after the first operation for sepsis caused by carbapenem-resistant *Klebsiella pneumoniae*.

4. Discussion

The patients with liver hydatid cysts should receive some form of treatment. Observation alone with ultrasound follow-up could be

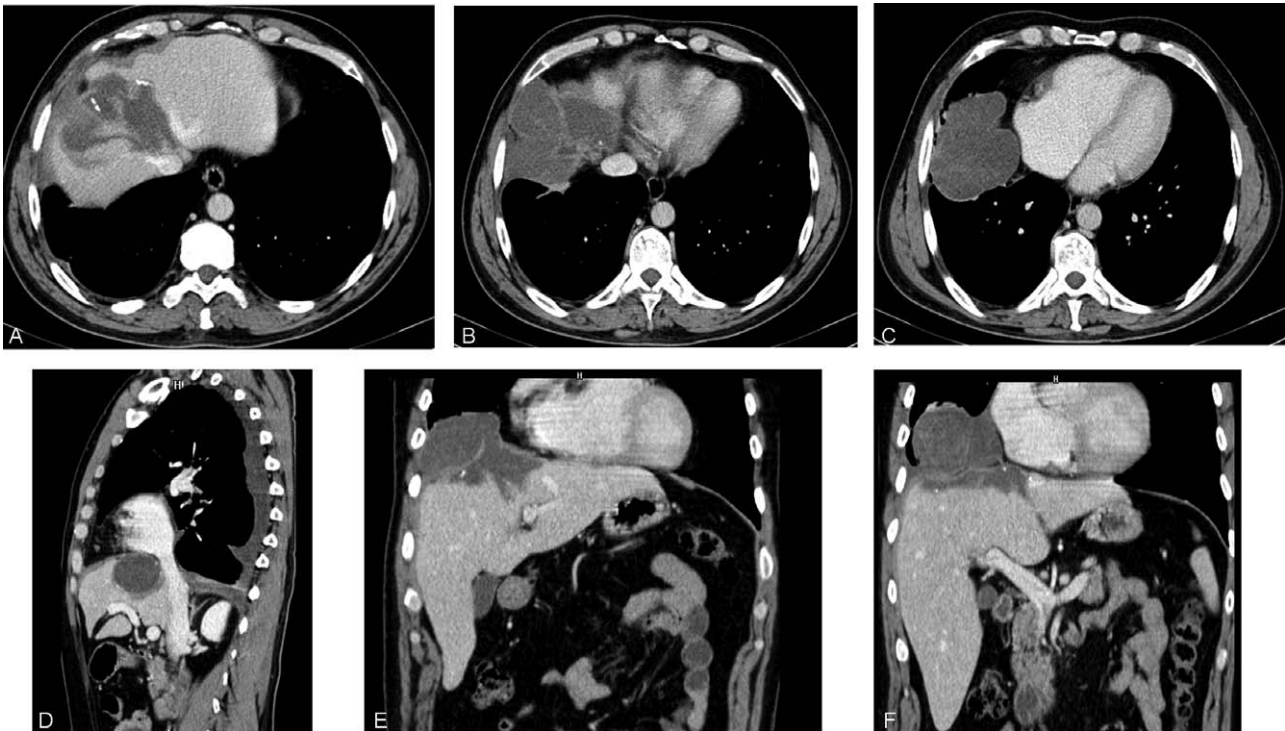


Figure 3. CT scan showing multiloculate hydatid cysts of the right hepatic lobe abutting the diaphragm and involving the lower pulmonary right lobe in a 44-year-old man. He underwent radical pericystectomy associated to diaphragm repair, and lower pulmonary lobectomy (A–C: CT axial view; D: CT lateral view; E and F: coronal view).

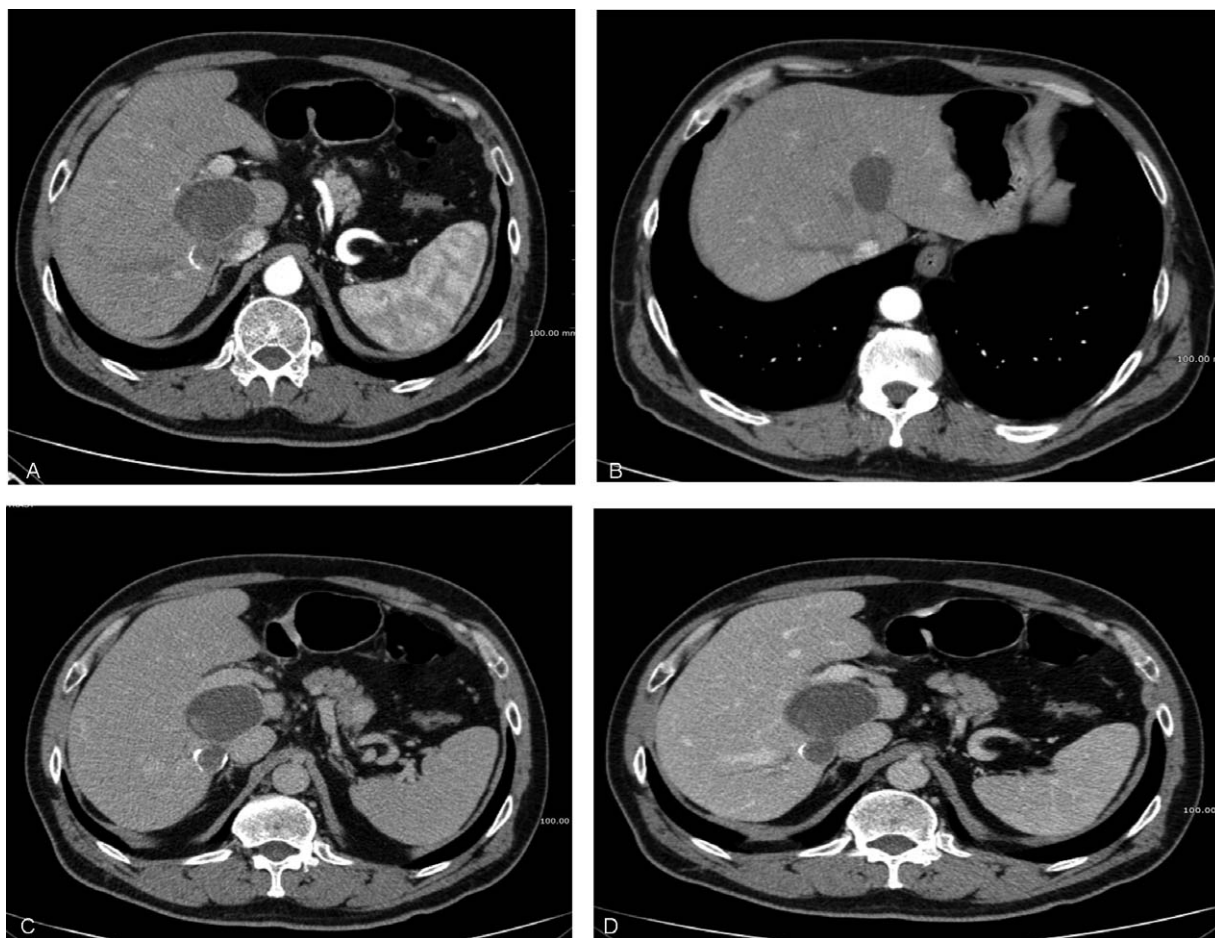


Figure 4. CT scan of a 68-year-old man with two hydatid cysts in close vicinity to the inferior vena cava. (A–D) A cysto-biliary communication was also present. Subtotal pericystectomy along with cholecystectomy and T-tube placement was performed.

only considered in selected patients with small and asymptomatic cysts containing dense calcifications, the latter being features of cyst death and inactivation.^[10] Surgery has been considered for years the mainstay of treatment. However, the management of hepatic hydatid disease has considerably changed over the last 20 years; in fact, the spreading of prevention programmes in endemic areas, along with the use of chemotherapy with benzimidazole compounds, has led to an effective control of the disease.^[3,9,11] In addition, advances in minimally invasive forms of treatment, such as PAIR (puncture–aspiration–injection–re-aspiration), have further reduced the need for surgical interventions.^[12,13] Nonetheless, surgery still maintains a prominent role when nonoperative management has failed or is not considered feasible, and also in complex and/or complicated hydatid cysts.

In surgical management of liver hydatidosis, different clinical situations may require more complex management than usual, however, there is no a unique definition of “complex” liver hydatidosis that is commonly accepted. According to our experience, we think that complex cases and complicated cases cannot be considered the same. A cyst can be complex, but not complicated (i.e., an uncomplicated giant cyst which requires complex management, or multiple cysts). In contrast, infected hydatid cysts, and those with cysto-biliary or cysto-bronchial communications, can be clearly considered as complicated cysts,

requiring a complex surgical procedure. Therefore, we arbitrarily created the aforementioned definition of “complex cases,” which refers to those cases which encompass complicated and non-complicated cysts that need difficult and skill-demanding surgical procedures. Gharbi’s and WHO classifications are commonly used in defining characteristics of hepatic cysts.^[14,15] However, none of them has utility in evaluating the complexity of management and the operative risk. Botrugno et al, in a cohort of 38 patients considered as complex hydatid cysts of liver those with a diameter ≥ 10 cm, as well as recurrent and multiple cysts,^[16] while others reported as complex those cases of difficult management.^[8,17,18]

In our series, 19% of the patients had a cysto-biliary communication, a common complication of hepatic echinococcosis reported in 26% to 80% of cases.^[4,11,19] Cysto-biliary communication generally causes severe symptoms due to biliary infection, but can present with a wide spectrum of clinical manifestations. Depending on the size and location of cysts, patients can be asymptomatic or have jaundice, cholangitis, liver abscess, and sepsis.^[6,19] Cysto-biliary communications can be diagnosed preoperatively or at the time of operation; in both cases they must be meticulously repaired.^[6] The cysts located in the central liver segments, multivesicular cysts, and those over 10 cm in diameter are more likely to fistulize with the biliary tree.^[2,6,19] Fistulization can occur in small-calibre ducts or major

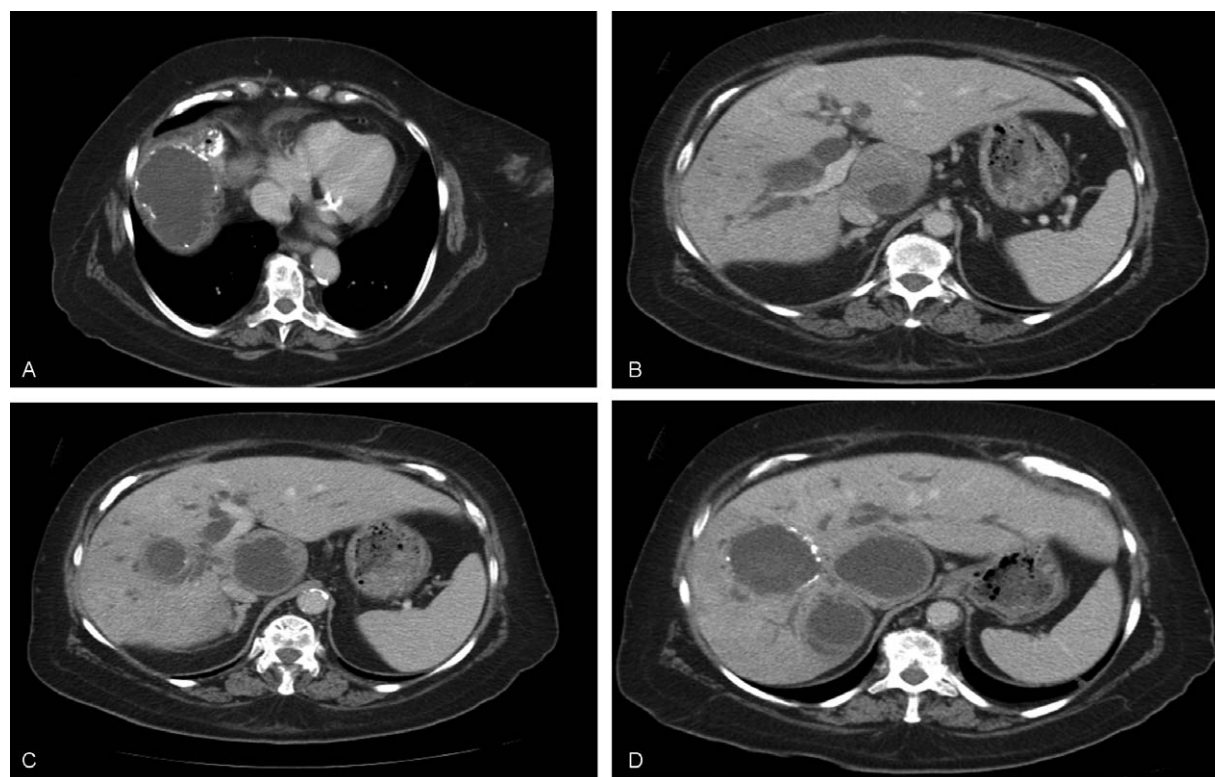


Figure 5. CT scan showing multiple hydatid cysts located in the right lobe of the liver in a 72-year-old man (A–D). There was a cysto-biliary communication with presence of hydatid material in the common bile duct. ERCP plus sphincterotomy was carried out before the surgical operation.

biliary ducts. In presence of minor leakages we used simple suture repair during total or partial pericystectomy, whereas a T-tube was placed in the common bile duct in presence of major biliary communications. It should be noted that cysto-biliary communications have been considered an important predictive factor for postoperative complications.^[5] In fact, the intrabiliary rupture of the cyst increases postoperative morbidity, ranging from 16% to 55%, and mortality, ranging from 1.25% and 7%.^[5,6,20]

Four patients of the present series underwent surgery for erosion of the diaphragm by the cyst wall, and in two of them there was also a communication with the bronchial tree. Thoracic complications of hepatic hydatid cysts are seen in ~2% to 16% of cases.^[21,22] Liver cyst can be infected and produce a secondary hepatic abscess that may rupture into a bronchus, or can be in direct communication with the bronchial tree. The clinical picture is predominately pulmonary, with respiratory symptoms such as

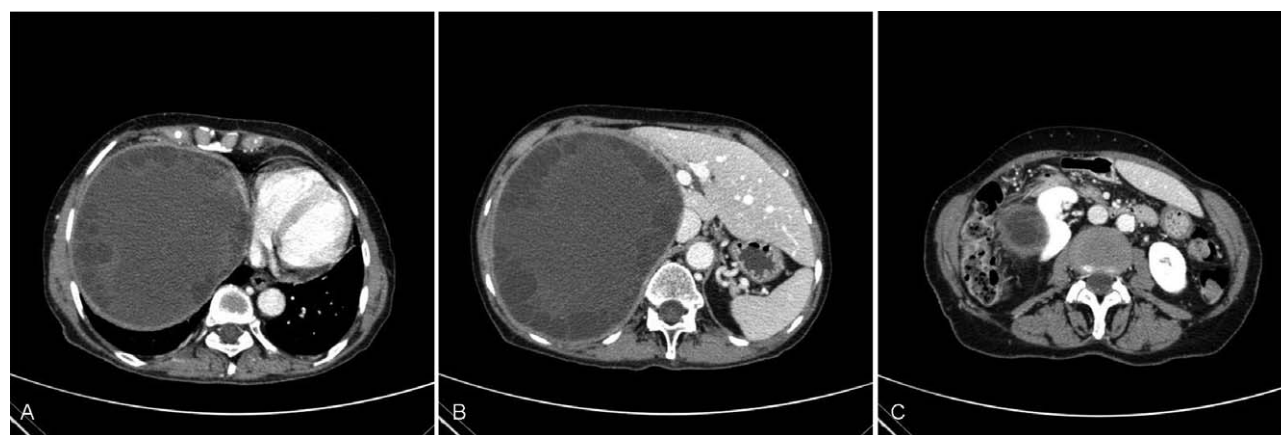


Figure 6. CT scan of a 75-year-old woman with a giant hydatid cyst measuring 25 cm in its major diameter containing daughter cysts, and causing mediastinal shift with displacement of the heart, inferior vena cava, stomach, and right kidney (A–C). To note, there was a compensatory hypertrophy of the left hepatic lobe (B). Operation performed: Right hepatectomy with diaphragm repair.

Table 4**Surgical treatment.**

	All patients (n = 55)	Complex Group (n = 42)	Non-Complex Group (n = 13)	P
Surgical procedure				<.001
Subtotal pericystectomy	5 (9.1%)	6 (14.3%)	0 (0%)	
Partial pericystectomy	12 (21.7%)	10 (23.8%)	1 (7.7%)	
Radical Pericystectomy	11 (20.0%)	2 (4.8%)	9 (69.2%)	
Hepatic segmentectomy	21 (38.2%)	19 (45.2%)	2 (15.4%)	
Right hepatectomy	3 (5.5%)	3 (7.1%)	0 (0%)	
Left hepatectomy	3 (5.5%)	2 (4.8%)	1 (7.7%)	
Additional procedure	35 (63.6%)	41 (97.6%)	8 (61.5%)	.02
Diaphragm repair	4 (11.4%)	4	0	
T-tube placement in the CBD	11 (31.4%)	10	1	
Cholecystectomy	15* (42.8%)	9	6	
Atypical lung resection	1 (1.8%)	1	0	
Right basal lobectomy	1 (2.8%)	1	0	
Bilio-enteric anastomosis (Roux-en-y)	1 (2.8%)	1	0	
Hysteroansectomy	1 (2.8%)	1	0	
Right Adrenalectomy†	1 (2.8%)	0	1	

CBD = Common Bile Duct.

* Excluding those cases in which an upfront right hepatectomy was carried out.

† Concomitant intervention for right adrenal incidentaloma.

cough, dyspnea, thoracic pain, biliptysis, or even respiratory failure.^[21,23] Patients with biliary-bronchial communications always require a complex procedure for their treatment, due to the contemporaneous involvement of liver, diaphragm, and lung. Although treatment of the hydatid liver cyst and diaphragm repair might be sufficient in some cases, as the pulmonary infection may resolve after the abolition of the biliary-bronchial communication,^[23,24] it is our opinion that also resection of the involved lung parenchyma should be warranted. In this series, a right lower lobectomy and an atypical pulmonary resection were carried out along with the surgical treatment of the liver cyst in two cases, through a thoracoabdominal incision. In one of them (described in Supplementary Material: videoclip 1, <http://links.lww.com/MD/F294>), also a costal resection was necessary due to inflammatory involvement of the chest wall. All these thoracoabdominal procedures were carried out by the same surgical team.

We have considered as complex cases those in which the cysts are in contact with or cause compression of major vascular structures. We observed these features in 30.9% of patients, most of whom were asymptomatic. Complete or incomplete obstruction of the portal vein or its main branches may result into decreased portal vein inflow as well as hepatic morphological and functional changes, including atrophy of the involved lobe and subsequent compensatory hypertrophy of the contralateral lobe.^[25,26] The position of the cyst represents an important factor influencing the choice of the surgical treatment. When the wall of the cyst is located either in direct contact with or in the close vicinity of major vessels such as vena cava, hepatic veins, portal vein, and/or main portal branches, the surgical treatment of the cyst is associated with an increased risk of intraoperative and postoperative bleeding.^[27] There is little evidence about those cases in the literature, with mostly case reports. In line with other authors,^[5] we usually prefer to perform a subtotal or a partial

Table 5**Postoperative complications.**

	All patients (n = 55)	Complex Group (n = 42)	Non-Complex Group (n = 13)	P
Total no. of patients with morbidity	15 (27.3%)	14 (33.3%)	1 (7.7%)	.07
Respiratory infection	1 (5.9%)	1 (6.7%)	0 (0%)	
Pleural effusion	3 (17.6%)	2 (13.3%)	1 (50%)	
Wound infection	1 (5.9%)	1 (6.7%)	0 (0%)	
Biliary fistula	5 (29.5%)	5 (33.3%)	0 (0%)	
Intra-abdominal abscess	3 (17.6%)	2 (13.3%)	1 (50%)	
Postoperative bleeding	3 (17.6%)	3 (20.0%)	0 (0%)	
Colonic ischemia	1 (5.9%)	1 (6.7%)	0 (0%)	
Total no. of complications*	17 (100%)	15 (100%)	2 (100%)	.14
Reoperation	4 (7.3%)	4 (9.5%)	0 (0%)	.24
30-day mortality	0 (0%)	0 (%)	0 (0%)	
1-year mortality	1 (1.8%)	1 (2.4%)	0 (0%)	.76
Median hospital stay (days, range)	10 (4–57)	12.0 (6–57)	7.0 (4–11)	<.001

* Four patients presented more than one complication.

pericystectomy in cases in whom extra-hepatic vessels are involved, leaving a patch of cystic wall attached to the vascular structure in order to minimize the risk of bleeding.

Most primary cases of liver hydatid cyst infection consist of a single cyst, but about 20% to 40% of infected people have multiple cysts.^[1] As mentioned above, the location of the cyst is important in choosing the most appropriate surgical intervention. The right lobe is the most commonly affected and the involvement of both lobes is not a frequent condition.^[6,16,28] However, patients with multiple and bilobar cysts have a major risk of intrabiliary rupture, as shown by Akcan et al.^[28] In patients with multiple bilobar cysts, who accounted for 11.9% of the present series, we performed radical or partial pericystectomies. Anatomical or non-anatomical liver resections are not warranted in these complex situations due to the risk of functional insufficiency of the liver remnant.

More than half of the patients included in our series (54.5%) had at least one hydatid cyst of the liver measuring 10 cm or more in diameter. While some authors defined cysts having that size as “giant” cysts,^[4,16] we rather prefer to consider as giant cysts those having a diameter of at least 15 cm. Those cysts can compress adjacent organs or structures and can be associated with complications. In particular, large cyst size increases the risk of rupture into the peritoneal cavity and hollow viscera.^[20,28,29,30] In the case series described herein, we operated on 6 patients with cysts having diameter superior to 15 cm. Surprisingly, all of them were asymptomatic or paucisymptomatic.

Surgical treatment of hydatid cysts of the liver traditionally encompasses conservative and radical interventions. Conservative operations, such as cystectomy, removal of cystic material, deroofting, and marsupialization procedures, mostly consist in drainage and evacuation of the cystic content.^[27] Radical approaches such as total, subtotal, or partial pericystectomy have as a goal the removal of the cyst with a rim of surrounding liver parenchyma.^[1,2,5,28,31] The ideal surgical treatment of liver hydatid cysts remains a matter of debate.^[12,32] In a recent meta-analysis, Pang et al demonstrated that radical procedures reduce the rates of postoperative complications and cyst recurrence when compared to conservative surgery.^[33] Although prospective studies comparing conservative vs radical approaches have not been done, we believe that radical surgical treatment should be always pursued, when feasible. In fact, many studies confirm that total removal of the cysts along with the entire pericystium is one of the more important issues in management of liver hydatidosis.^[2,18] It should be noted that recurrence rates after surgical treatment vary from 1% to 20%, even if they are lower than 5% in experienced centers.^[4] To note, 10 of the patients described herein underwent surgery for recurrent hydatid cyst of the liver. Surgical treatment was significantly different between the two groups of our study (Complex vs non-Complex), due to the fact that radical pericystectomy was the most performed operation in the non-complex group, while it was carried out in only 4.8% of patients in the Complex group. In the latter, due to the location and characteristics of the cysts, we performed more often partial or sub-total pericystectomies, or liver resections. In particular, in 6 out of 55 patients of our experience, a major hepatic resection, such as right or left hepatectomy was necessary due to the extensive liver involvement. In a recent report, Ramia et al performed major hepatectomy in cases of occupation of an entire lobe, extreme biliary or vascular involvement, or recurrent cysts.^[32] Reoperation rate for postoperative complications in the

present series was 7.3% in the entire cohort. This finding was consistent with other studies such as Secchi et al, who reported a reoperation rate of 9.6% in a multicentre series of 1412 patients.^[12] With regards to additional procedures other than hydatid cyst treatment, we observed that they were more frequently necessary for the complete treatment of the disease in the Complex group ($P=.02$). These concomitant operations added to the complexity of surgical operations and probably had a role in the occurrence and severity of postoperative complications.

The literature suggests that laparoscopic treatment can be used in selected patients with benign and malignant liver diseases.^[34] Laparoscopy treatment of cystic liver hydatidosis has been reserved for small and uncomplicated cysts close to the liver surface.^[3,8] We used that approach in one patient belonging to the no-Complex Group in whom the risk of cyst rupture during the operation was deemed to be very low, based on the preoperative work-up. As for patients presenting with complicated hydatid cysts, there is a common agreement that they do require open liver surgery.^[8]

To note, 27% of patients in our series developed postoperative morbidity. This finding is consistent with previous studies. Abuoglu et al in a cohort of 40 patients operated on for hydatid cysts larger than 10 cm reported postoperative abdominal complications in 22.5% of cases.^[4] Akcan et al reported postoperative complication rates of 27% in patients operated on for complicated hydatid cysts.^[28] Similarly, Georgiou et al observed a complication rate of 29.3% after surgical conservative techniques.^[2] In the cohort published by Botrugno et al, which included patients who underwent surgery for complex hydatid cysts of the liver, overall morbidity was 26.3%.^[16] As expected, complications were more frequent in the Complex group, although not in a significant manner. One of the most dreaded complications in surgical treatment of hydatid cyst, and in liver resection surgery in general, is the formation of biliary fistulas. Fortunately, most of them spontaneously close or are being successfully treated with ERCP and sphincteroplasty.^[2,5,35] In the present series, only in one patient belonging to the Complex Group a re-laparotomy was necessary for persistent biliary fistula.

In accordance with others, it is our thought that surgical treatment of hydatid cysts will be more frequently reserved for the most complex and skill-demanding cases, thus patients should be preferably referred to centers experienced not only in hepatobiliary surgery, but also in management of hydatid disease. A definition of complex cases may also be useful in stratifying patients for surgical treatment.

We recognize that the present study has some limitations, the main being its retrospective design and the small sample size. However, there are few studies in the literature that have described special challenging cases, in which the management of complicated cysts has been reported together with that of complex albeit paucisymptomatic cases.

5. Conclusions

Management of liver cystic hydatidosis may represent a challenging situation for hepatic surgeons, burdened by serious complications. Cysts of large diameter (>15 cm), or compressing main vascular structures, or located in both hemilivers, albeit often of little clinical relevance, can be considered as complex and skill-demanding cases, in the same ways as complicated hydatid cysts.

Author contributions

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