Development in diagnosis and treatment of hepatic echinococcosis in a surgical department of a mediterranean centre over a 20-years period

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Abstract

Liver echinococcosis is an endemic disease in some areas of the world like Middle East and is a serious problem both from social-economic and clinical-therapeutical stand-points in other areas like Mediterranean regions. We report our experience on hepatic echinococcosis diagnosis and treatment over a 20-year period. In this retrospective study we have reviewed 89 patients affected by hepatic hydatidosis who underwent surgery in our Unit (between November 1975 and October 1995). Patients were divided into two grou - ps: group A including 65 patients (30 males and 35 fema - les) operated on between 1975 and 1988 and group B including 24 patients (11 males and 13 females) operated on between 1989 and 1995. Main outcome measures repor - ted were recurrence of echinococcosis, association with sur - gical procedure, complications, diagnostic trial.
In group A we performed 41 marsupializations, 20 sub- total pericystectomies and 4 total pericystectomies; while 14 total pericystectomies and 10 liver resections were performed in group B. Postoperative hospital stay was of 30 days for group A patients and of 18 days for group B. A single death occured in group A. Recurrences occured in group A only (11 cases equal to 17%) and were treated with total pericystectomies (8 cases) and liver resections (3 cases).
Our study shows the development of diagnostic procedures trial during the 20-year period and the different surgical approaches between the two groups.
Nevertheless we believe that surgical procedures should be tailored to each patient avoid H high surgical risk due to the benign nature of the disease.
Key words: Hepatic echinococcosis, hydatidosis, liver surgery.

Introduction

The liver is the organ most often affected by hydatid disease, hepatic involvement occuring in about 70% of cases (1); it is the main organ where the Esacanto embryon gets intravenously and stops due to very small portal capillaries and to a possible presence of parasite organotropism.
Echinococcosis, an endemic disease affecting some areas of the world, is the most frequent cause of cysts formation in the liver (2).
It is a well-known pathology since a long time, its biological cycle being described for the first time in 1862 (3) and its geographic distribution recently modified.
Its incidence appears to be constantly increasing in some areas such as the United States.
It was mainly due to migration from those areas, where hepatic hydatidosis is considered an endemic pathology. On the other hand, in some areas like Sicily that, due to hydatidosis, are traditionally considered endemic, the frequency of this pathology had a decrease in the last years thanks to a better prophylaxis of cattle and to a remarkable decrease in pastoral activity.
Nevertheless, hepatic hydatidosis is a very serious problem both from social-economic and clinical-therapeutical standpoints.
During these 20 years remarkable improvements were achieved in diagnostic imaging techniques of hepatic echinococcosis.

Surgery is still the treatment of choice in hepatic echinococcosis although the trend to perform radical rather than conservative procedures has been the subject of some controversy.
Efficacy of anti-helminthic drugs or recently proposed mini-invasive surgical procedures are questionable (4).
This paper reports our experience on diagnosis and treatment of this pathology developed over the last 20 years.

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Tab. I – TREATED PATIENTS

	Males		Females	
	N.	%	N.	%
Group A (mean age 44 years, range 14-75)	30	46	35	54
Group B (mean age 42 years, range 13-70)	11	47	15	53

Materials and methods

Between November 1975 and October 1995 we performed surgery on 89 patients affected by hepatic hydatidosis in our Unit.
In order to better understand the evolution of diagnostic approaches and therapeutical attitudes over this period, we have divided our patients into two groups: Group A and B.
Group A included 65 patients who underwent surgery between 1975 and 1988, 46% of them were male and 54% were female with an age range of 14 to 75 years.
Group B included 24 patients who underwent surgery between 1989 and 1995, 47% of them were male and 53% female with an age range of 13 to 70 years (Tab. I).
Group A consisted of 57 patients (87.6%) with hepatic cysts, 7 patients with hepatopulmonary extension, and only one with cysts located in the liver and spleen.
In Group B, 92% of patients had disease in the liver only and 8% in the liver and lung. In both groups the distribution in the right lobe of the liver was higher than in the left one (66% vs 34% in Group A, and 68% vs 32% in Group B).
In both groups cysts were solitary in 85% and multiple in 15% with a size ranging from 3 to 15 cm in diameter. The first group included 2 recurrences coming from other Units, the second one included 9 recurrences previously treated in other Units.
In particular, Group A revealed a slighter different symptomatology compared to Group B. Hepatomegaly occured in 70% and 55% of Group A and B respectively. Palpable abdomen mass occured in 63% (Group A) and 47% (Group B). Both groups didn't showed any substantial differences in the frequency of other symptoms such as fever, pain and jaundice.
Remarkable differences were observed in diagnostic procedures performed in these groups.
In the first part of our experience, we always performed a plain abdominal Rx, liver scintiscan (49%), intravenous cholangiography in 28% of patients with suspected biliary tract involvement. In the most severe cases, we performed ultrasonography (45%), CT scan (33%) or flebocavography of extrahepatic veins (12%). Ultrasonography and Ct scan were performed in all Group. B patients. We have performed 5 arteriographies for anatomic-surgical investigation. Endoscopic retrograde cholangiopancreatography (ERPC) was performed in 4 patients with suspected biliary tract involvement (Tab. II). Test for hematic eosinophilia and the Casoni test were always performed in Group A patients. On the contrary, in Group B we followed routinerely the Elisa method for the antibody assay and in the follow-up we introduced use of the RAST test.
We always evaluated the hepatic function before surgery. When we planned to perform liver resection in Group. B patients or in presence of large cysts substituting the parenchyma, we carried out MEGX (monoethylglycinexilydide) test to evaluate the hepatic functional reserve.
Intraoperative ultrasonography was performed in all Group. B patients at surgery.
Incision was performed in Group A according to location and size of the lesion; we performed a thoracophrenolaparotomy in 7 patients with hepatopulmonary extension, a right subcostal incision in 27 patients affected by hepatic hydatid disease in the right lobe. We performed a bilateral subcostal resection sometimes extended to the sternum in 31 patients with large cysts located in the right lobe, or confined in the liver and spleen, or in the inner part of the liver or in the left lobe. We performed a thoracolaparotomy in 2 patients with hepatopulmonary extension (Group B). We performed a right subcostal incision extended down to the 12th rib and up to the ensiform apophysis of the sternum in patients with cysts located in the liver only aside from involved lobes and cyst size.
As scolicidal agent, we first administered formaldehyde solution, then hypertonic solution of sodium chloride and at last hydrogen peroxide.
Sub-total pericystectomy refers to the removal of the pericystium,leaving in situ areas adherent to important

Tab. II – DIAGNOSTIQUE PROCEDURES

Group A		*Group B*	
Abdominal RX	100%	Ultrasonography	100%
Hepatic scintigraphy	49%	Ct scan	100%
Endovenous cholangiography	28%	Arteriography	21%
Flebocavography of extrahepatic veins	12%	ERCP	17%
Ultrasonography	95%		
Ctscan	33%		

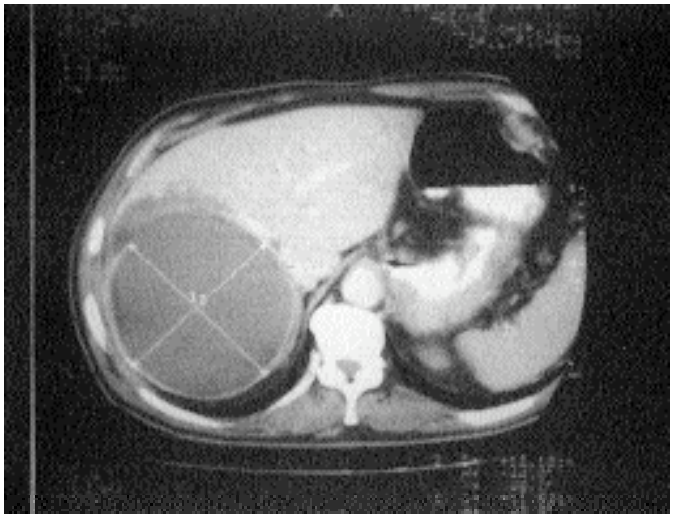

Fig. 1: Sub-total pericystectomy for hydatid liver cyst. Pre operative CT: the cyst occupies the entire right lobe of the liver.

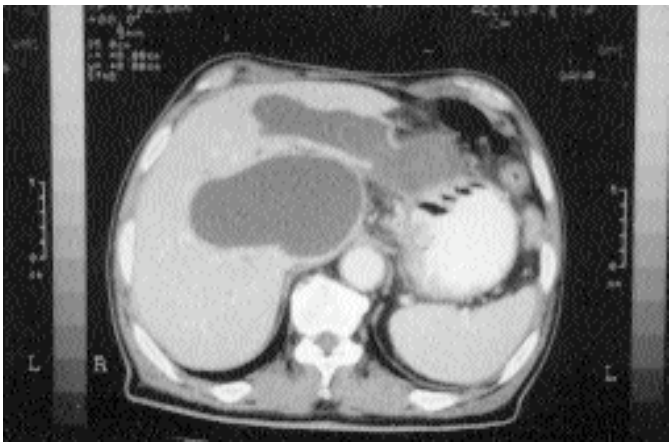

Fig. 2: Multiple hydatid cysts localized in the left lateral segment and in the caudate lobe. Left lobectomy and subtotal pericystecomy for the cyst of the caudate lobe.

vascular structures and is indicated in the case of large cyst with extended intrahepatic development (Fig. 1).
Liver resections are restricted to selected cases of single large or multiple cysts destroying a segment or an entire lobe (Fig. 2).
Hepatic infestation with E. granulous was histologically confirmed in all cases. Mebendazole was administered in Group B patients, 50mg/kg daily for 15 days before surgery and at least 3 months after.
All patients were invited to periodic control sessions at which time laboratory tests, hepatic ultrasonography and chest x-ray were performed. Last control was in 1997 (mean control period: 5 years).

Results

We performed 49 marsupializations, 20 subtotal pericystectomies and 4 total pericystectomies in Group A, while 14 total pericystectomies and 10 liver resections (Tab. III) were carried out in Group B.

Tab. III – SURGICAL PROCEDURES

	Group A		*Group B*	
	N.	*%*	*N.*	*%*
Marsupialization	41	63	–	–
Sub-total pericystectomies	20	3	–	–
Total pericystectomies	4	6	14	58
Liver resection	–	–	10	42

Tab. IV – POSTOPERATIVE COMPLICATIONS IN THE TWO GROUPS

	Group A	*Group B*
Biliary fistula	7	–
Transient choleragie	4	–
Abscesses of residual cavity	2	2
Subphrenic abscesses	3	–
Pleural drainage	2	1
Intrabiliary rupture of the cyst	1	–
Sclerosing cholangitis	1	–

Among radical procedures, we performed 14 "closed" total pericystectomies, 4 "open" total pericystectomies and 10 liver resections of which 7 wedge resections, 1 left lobectomy and 2 regulated left hepatectomy.
Surgery was the treatment of choice in both groups, preoperative diagnosis being correctly performed.
There was no accidental echinococcosis recurrence during laparotomies carried out for other diseases.
Concomitant biliary pathology was evidenced in 12 (Group A) and in 4 cases (Group B). Suspected biliary tracts involvement was suggested by symptomatology (10 cases) or by preoperative diagnosis (4 cases) or by demonstration of abundant biliary contents within the cyst cavity at surgery.
Postoperative hospital stay was of 30 and 18 days for Group A and B respectively.
One Group A patient presenting with severe episodes of acute cholangitis, due to intrabiliary rupture of the cyst, died of hepatorenal failure. There was no mortality in Group B.
We observed a 30% rate of postoperative complications in our first Group: biliary fistula (7 cases), transient choleragie (4 cases), abscesses of residual cavity (2 cases), subphrenic abscesses (3 cases), pleural effusions (2 cases), sclerosing cholangitis (1 case), intrabiliary rupture of the cyst (1 case). The second group rate of complications was 12%: one case of pleural effusion and 2 cases of abscesses of residual cavity (Tab. IV). We recorded 11 recurrences treated with total pericystectomy (7 cases); with sub-total perycistectomy (2 cases); and liver resection (2 cases).

Discussion

Objective of this study is to review our experience on the evolution of diagnostic and therapeutical strategy of hepatic echinococcosis over a 20-year period.
We rarely observed changes in laboratory tests (6) (7) in this disease even though hepatic infestation with E. Granoulous causes an IgE production.
We agree with other authors (2) (8) that the utility of a serological diagnosis in the definition of hepatic echinococcosis is questionable, except when diagnostique techniques may help differentiate hydatid cysts from abscesses or infestations with other parasite. In the first period of our experience, we routinely used the Casoni test which was useless due its non specificity. Consequently, we introduced the Elisa method for specific antibody assay. Despite this method was able to differentiate antibodies of the previous disease from those of the ongoing one, we haven't recorded satisfactory results.
We believe that introducing laboratory tests in the follow-up of treated patients could represent an innovative aspect aiming at performing an early diagnosis of recurrence, as we can see from our second group of patients being evaluated with the RAST test.
Ultrasonography and CT scan are commonly preferred to traditional radiography and scintigraphy no more used. Ultrasonography shows the morphological characteristic of the cyst, possible presence of scolices inside, calcificification of the cyst wall and involvement of the biliary tree. Besides, in association with Doppler, it is possible to study venous and arterial vessels feeding the liver that could be included in the formation of cysts (9) (10) (11). CT scan can be useful in doubtful cases (12) and can provide information. We performed arteriography only for anatomic-surgical investigation in few selected patients. The Magnetic Resonance Imaging (MRI) can reveal the communication with suprahepatic vessels and vena cava. In order to confirm preoperatively a communication between cysts and biliary tree, endovenous cholangiography was used in the earlier part of our experience and was replaced later by ERPC which proved to be very useful in case of complications. The endoscopic procedure was also helpful in post-operative patients, showing an external biliary fistula; we routinely peform endoscopic sphincterotomy in patient with externa biliary fistula or in patient with liver hydatid cysts that have ruptured into the biliary tree. The role of Magnetic Resonance Cholangiography in evaluating inflammotory stenosis of the bile duct related to liver ydatidosis is comparable to that of endoscopic cholangiography (Fig. 3), but doesn't has the advantage to allowing any therapy at the time of initial diagnosis (13).
Although surgery is unanimously considered the treatment of choice for hepatic hydatidosis, controversies still exist about the use of conservative or radical procedures. In the past the sole therapy to be used was conservative surgical management, because was used to be thought that drainege of the cyst with iodine or formalin solution would ensure against the risk of recurrence (14) (15). This belief was rejected when the high recurrence rate among patients treated in the above manner was shown. Today, as general rule, radical surgery is prefered to conservative surgery, mainly because of certain physiopathological consideration, of which exogenous vesciculation is the most important (16) (17).
In the first part of our experience we performed almost only conservative procedures removing the endocyst and cyst contents but not the pericyst. The residual cavity was managed either by omentoplasty or by marsupializations. Surgery should aim at removing the parasite exeresis of cystic wall with evacuation of residual cavity and prevention of complications and recurrences. Radical procedures are associated with a lower rate of postoperative complications (mainly biliary fistula) and recurrence, because treatment of leaks in the residual cavity by direct binding is possible. Total pericystectomy is, theoretically, the ideal treatment for liver ydatidosis; complete exicision of the pericystium favors the healing

Fig. 3: Endoscopic colangiography: stenosis of the common bile duct following sclerosis related to ydatidic sludge.

process of the remaning cavity (18). Nevertheless, radical procedures such as total pericystectomies may lead to a high incidence of intraoperative bleeding in particular in cases of suspected communication between the pericyst and suprahepatic vessels or vena cava.
Some authors consider radical procedures such as resection inappropriate claiming that the risk of damaging a functional parenchyma may be too high for a benign disease (19). Although conservative procedures imply low surgical risks, they are associated to a high rate of recurrences and postoperative complications (20) (21). We agree with other authors (12) (22) (23) (24) that the surgical treatment should be tailored to the individual patient. Furthermore radical surgery is able to clear scolicidal agents such as formaldehyde solution, hypertonic saline solution and hydrogen peroxide which are reported to be the primary cause of severe chemical cholangitis (3) (10) (25). The current trend to perform radical surgery is highly supported by the use of intraoperative ultrasonography showing special anatomical features of the liver due to the progressive growth of the parasite that leaves vascular structures to penetrate into the biliary system. Biliary leakage is known to be the most common postoperative complication following surgery for hydatid liver disease. It is considered the "weak point" of conservative surgery (26) (20). Also in our series bilary leakage represented the most common complication: we had 8 cases in group A end none in group B. We had a single death due to intrabiliary rupture of the cyst. In the past, in case of Hydatid discharge into the biliary tract, we performed a surgical exploration of the common bile duct and subsequently a choledoduodenostomy. Afterwards, we performed ERCP and a papillosphincterotomy to treat these complications. A further aspect of our analysis was related to recurrences that in our experience always occured in the same site of the first surgery and only after conservative procedures. As many authors claim (27) (28) (29) the best treatment for local recurrence due to exogenous vesciculation is total pericystectomy or subtotal pericystectomy next to main vessels. Recent studies have evaluated the efficacy of anti-helminthic drugs on hidatid disease. There are many authors (30) (31) (32) (33) claiming that this kind of drugs reduces hydatic sludge and liquid together with endocystic pressure. On the other hand, there are authors (34) (35) (36) (37) who do not believe in the efficacy of this treatment. We believe that surgery could not be replaced by pharmacological therapy; antihelminthic drugs should be associated before and after the surgery in our opinion.
In conclusion, we feel that surgical strategy should be tailored to each patient avoiding high surgical risks due to the benign nature of the disease; an early diagnosis is essential for the rigth treatment because the latency period between infestation and presentation can exclude the patient from a radical cure. Furthermore a careful preoperative work-up is the key to have a good surgical indication which depend on patient's fitness and on anatomical accessibility of the cyst.
There still exist controversies about the use of laparoscopy (38) or injection-guided aspiration (39) (4) due to the lack of statistically significant data.

Riassunto

In questo studio retrospettivo abbiamo voluto rivedere la nostra esperienza con l'echinococcosi epatica nel corso degli ultimi vent'anni, in riferimento all'evoluzione della sua diagnosi e del suo trattamento.
I pazienti sono stati suddivisi in due gruppi: il gruppo A che racchiude 65 pazienti (30 maschi e 35 femmine) operati nel periodo compreso tra il 1973 ed il 1988, ed il gruppo B che comprende 24 pazienti (11 maschi e 13 femmine) operati nel periodo compreso tra il 1989 ed il 1993. Nel gruppo A sono state eseguite 41 tunnellizzazioni e marsupializzazioni, 20 pericistectomie parziali e 4 pericistectomie totali. Nel gruppo B sono state eseguite 14 pericistectomie totali e 10 resezioni epatiche. La degenza post-operatoria è stata di 30 giorni per i pazienti del gruppo A e di 18 per quelli del gruppo B. Un solo decesso si è verificato nel gruppo A, nessuno nel gruppo B. Le recidive sono state tutte a carico del gruppo A (11 casi pari al 17 %) e sono state trattate in 8 casi con reinterventi di pericistectomia totale, ed in 3 casi con resezione epatica.
Sebbene non esistano differenze in termini di mortalità fra i due gruppi della nostra serie per quanto riguarda la degenza media postoperatoria, l'insorgenza di complicanze post-operatorie e le recidive, sono evidenti dei miglioramenti correlati ad un approccio chirurgico più radicale.

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Commento Commentary

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In questo studio clinico retrospettivo gli autori hanno rivisitato la loro esperienza con l'idatidosi epatica in un periodo di tempo di 20 anni (1975-1995). Vengono analizzati i cambiamenti, realizzati nel corso di questo ventennio, dell'iter diagnostico e dell'approccio terapeutico di questa patologia. In particolare, attraverso lo studio dei due gruppi di pazienti, si evince come con il progredire della diagnostica nel campo della patologia epatobiliare e dell'affinamento della chirurgia epatica sia cambiato l'impatto con l'echinococcosi epatica. Infatti l'ecografia e la TAC rappresentano ormai i punti cardine della diagnostica preoperatoria e la ERCP risulta sempre più importante nella diagnosi e nell'eventuale trattamento di complicanze legate al coinvolgimento dell'albero biliare.
La diagnostica sierologica si è arricchita di nuove metodiche e trova una sua valida collocazione anche nel follow-up di questi pazienti. Per quanto riguarda la terapia, sebbene pare, dall'analisi del lavoro, che ci sia una tendenza sempre più marcata verso la realizzzazione di interventi radicali, mi sembra che la considerazione più importante sia quello espressa dagli autori nel finale del lavoro in cui affermano che, trattandosi di patologia benigna, l'indicazione alla terapia chirurgica radicale debba sempre essere pesata con molta attenzione valutando rischi e benefici. Condivido la loro conclusione che non esiste una terapia standardizzata per il trattamento di questa patologia ma ogni intervento deve essere tagliato su misura per ogni singolo paziente.

In this retrospective study the authors report their experience with liver echinococcosis diagnosis and treatment over a 20-year period. The study shows the development of diagnostic procedures during the 20-year period and the different surgical approches between the two groups analyzed. The impact with liver idatidosis has changed in a surgical department. Ultrasonography and CT scan represent the main tools of the preoperative imaging and ERCP plays a first role in the diagnosis and treatment of the complications related to liver echinococcosis. Serological diagnosis could be usefull particulary in the follow-up of trated patients to detect early recurrence. Although the number of radical procedures is bigger in the second group than in the first one I agree with the authors that surgery should be tailored to each patient avoiding high surgical risk due the benign nature of the disease.

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