

Treatment of complications of hepatic hydatid disease by ERCP: our experience



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C. Sciumè, G. Geraci, F. Pisello, F. Li Volsi,
T. Facella, G. Modica

Università degli Studi di Palermo
Azienda Ospedaliero-Universitaria Policlinico
"Paolo Giaccone"
Dipartimento di Chirurgia Generale d'Urgenza
e dei Trapianti d'Organo
Sezione di Chirurgia Generale ad Indirizzo Toracico
Direttore: Prof. Giuseppe Modica

Introduction

Liver hydatidosis is a widespread parasitic infection caused by the tapeworm *Echinococcus granulosus*. The disease is endemic in some cattle and sheep-herding areas of the world, and is prevalent in Mediterranean countries such as Italy (endemic in Sardinia with 14.32 cases over 100000 people and Sicily with 4.5/100000) (1). The liver is the most frequently involved organ (60%-74%) with 60% to 85% of the cysts located in the right lobe. The disease usually does not cause symptoms, but cyst rupture into the biliary tree is a common complication (5%-25%) that can lead to obstructive jaundice, cholangitis, (or both) or acute pancreatitis (0.4%) when daughter cysts and fragmented membranes obstruct the biliary tree (2-3).

Surgery is the only radical and effective method indicated for patients with symptoms, for complications of hydatid disease, and for asymptomatic patients unresponsive to treatment with systemic antihelminthics (4) and the specific operative approach depends on the number and location of the cysts and associated complications. The most common complication of surgery is a fistula between the biliary tract and the residual cavity with persistent postoperative external drainage; another

Riassunto

TRATTAMENTO DELLE COMPLICANZE DELL'IDATIDOSI EPATICA MEDIANTE ERCP: NOSTRA ESPERIENZA.

Premessa: Lo scopo del nostro studio è quello di valutare l'efficacia della sfinterotomia endoscopica con il posizionamento di endoprotesi biliare o sondino naso-biliare davanti alle complicanze pre e postoperatorie che possono insorgere nella idatidosi epatica (fistole, compressioni, colestasi, franca rottura della cisti nella via biliare).

Materiali e metodi: Durante il periodo 1994-2003, 22 pazienti (12 uomini e 10 donne, età media 55.4 anni, range 16-65 anni) sono stati sottoposti a trattamento endoscopico per complicanze dell'idatidosi epatica. La indicazione ad eseguire la ERCP nei 5 pazienti con complicanze preoperatorie (Gruppo A) è stato l'ittero ostruttivo, associato a colangite acuta in 4 soggetti 4 (80%) e a pancreatite acuta in 1 (20%). Nei 17 pazienti In trattati per complicanze correlate alla cisti dopo l'intervento chirurgico (Gruppo B), l'indicazione è stata la colangite acuta in 6 (36%), l'ittero ostruttivo in 3 (i 7%), e una fistola biliare esterna in 8 pazienti (47%).

Risultati: Nel Gruppo A, sono stati rinvenuti detriti idatidei e cisti figlie in tutti i pazienti, sottoposti quindi ad ERCP e a toilette della VBP in assenza di complicanze maggiori legate alla procedura endoscopica. Negli 8 pazienti del Gruppo B affetti da fistula biliare esterna, dopo la ERCP si è assistito alla chiusura della fistula in 10-25 giorni (in 4 pazienti con fistola biliare a bassa portata è stato posizionato un sondino naso-biliare) o in 4-6 settimane (in 4 pazienti con fistola biliare ad alta portata è stata posizionata una endoprotesi biliare). In 218 pazienti (25%) con fistula biliare esterna è stata osservata anche una coledocolitiasi associata, che è stata trattata consensualmente per via endoscopica. In tutti i 9 pazienti con ittero ostruttivo o colangite postoperatoria, alla ERCP sono stati ritrovati detriti di cisti che ostruivano la via biliare. L'intervento chirurgico è stato evitato in un paziente del Gruppo A ed in tutti i pazienti del Gruppo B, e dopo il trattamento, tutti i pazienti sono rimasti asintomatici.

Conclusioni: La sfinterotomia endoscopia con posizionamento di sondino naso-biliare o di endoprotesi biliare è

un metodo sicuro ed efficace nel trattamento delle complicanze biliari pre e postoperatorie della idatidosi epatica.

Parole chiave: Idatidosi epatica complicata, echinococco, management, ERCP (colangiopancreatografia retrograda endoscopica).

postoperative complication is acute cholangitis and/or obstructive jaundice caused by hydatid remnants within the biliary tract.

Endoscopic methods are reportedly as effective alternative to surgery only for the treatment of such complications, with lower morbidity and reductions in hospital stay.

This article presents our experience with the endoscopic management of biliary complications of hepatic hydatid disease.

Patients and methods

During the period 1994-2003, a total 22 patients (12 male and 10 female, mean age 55.4 years, range 16-65 yrs) with hydatid diseases and biliary complications underwent ERCP and endoscopic sphincterotomy (ES). We divided the patients in two groups: in 5 patients who had not undergone surgery (**Group A**), the indication for ERCP was obstructive jaundice in all 5 patients, with acute cholangitis in 4 (80%) and acute pancreatitis in one (20%). Among 17 patients who had undergone surgery for hepatic hydatid cysts (**Group B**), the indications for ERCP were acute cholangitis in 6 (36%), obstructive jaundice in 3 (17%), and persistent external drainage in 8 patients (47%). Clinical features and laboratory data were collected before and after endoscopy (Table I).

The diagnosis of hydatid cyst in Group A patients was

based on US, CT, and serologic data (indirect immunofluorescence). ERCP and ES were carried out by using standard techniques, endoscopes, and accessories. Echinococcal material was removed with a balloon extraction catheter (EBL-1T-200; Wilson Cook Medical Inc., Winston-Salem, N.C.) or with Dormia basket (FG-22-Q-I; Olympus Italy, Milan).

After sphincterotomy, either a Dormia basket or extraction balloon catheters were used to remove the daughter vesicle material from the biliary tract.

Intravenous Levofloxacin was administered as a prophylactic measure 30 minutes before the procedure.

Results

Hydatid vesicles and membranes were evident within the biliary tract in all patients in Group A. They underwent ES and removal of the hydatid remnants or daughter-cysts. All 5 patients have remained asymptomatic during follow-up, which ranges from 1 to 5 years. Four of these patients subsequently underwent elective surgery for treatment of hydatid cysts after treating with albendazole 400 mg twice daily for 2 months with 4-week courses of therapy separated by intervals of 2 weeks without treatment (5). In 1 patients the treatment was nonsurgically by means of ERCP and adjuvant therapy (disappearance of cyst during one-year follow up) (25).

8 of the 17 patients in Group B presented with persistent external drainage caused by biliary fistula from the residual cavity. They underwent ES to decrease the pressure in the biliary tract, with positioning of naso-biliary drain in the 4 patients with low-flow fistula (400 ml/die) or with biliary endoprosthesis in the four patients with high-flow fistula (> 400 ml/die). We used plastic material with 10 Fr diameter. The healing of fistula was observed, respectively in 15-20 days (in the nasobiliary group) and in 4-6 weeks (in the endoprosthesis group).

Table I - INDICATIONS AND CHOLANGIOGRAPHIC FINDINGS IN THE 22 PATIENTS WITH COMPLICATED HEPATIC HYDATID DISEASE TREATED ENDOSCOPICALLY

Indications for ERCP (n)	Cholangiographic findings (n)
<i>Non-surgically treated patients (5/22 = 23%) = Group A</i>	
Obstructive jaundice (5) (4 with acute cholangitis and 1 with acute pancreatitis)	Biliary fistula (1 = 20%) Hydatid vesicles in the biliary tract (5 = 100%) Dilated biliary duct (5 = 100%)
<i>Post-surgically treated patients (17/22 = 77%) = Group B</i>	
Persistent external drainage (8 = 47%)	external biliary fistulas (8 = 100%) Associated choledocholithiasis (2 = 25%)
Obstructive jaundice with cholangitis (9 = 53%)	Cyst remnants in the biliary tract (9 = 100%) Associated choledocholithiasis (2 = 22%)

Table II - RESULTS

<i>Group A</i>	<i>5/5 = 100% → ERCP + ES + toilette biliary tree</i>	
Non-surgically treated patients	4/5 = 80% → underwent elective surgical treatment	
	1/5 = 20% → disappearance of cyst after ERCP and medical therapy	
<i>Group B</i>	<i>17/17 = 100% → ERCP + ES</i>	
Post-surgically treated patients	8/17 = 47% with external biliary fistula	<input type="checkbox"/> 4/8 low-flow fistula @ naso-biliary drainage (healing 15-20 days)
		<input type="checkbox"/> 4/8 high-flow fistula → biliary endoprosthesis (healing 4-6 weeks)
	(2/8 associated choledocholithiasis)	<input type="checkbox"/> ERCP + ES + toilette biliary tree
	9/17 = 53% with jaundice ± cholangitis	<input type="checkbox"/> 9/9 endoscopic toilette
	(2/19 associated choledocholithiasis)	<input type="checkbox"/> ERCP + ES + toilette biliary tree

In 2 of 8 patients (25%) with postoperative external biliary fistulas was observed associated choledocholithiasis: the toilette of VBP was achieved in all cases with Dormia basket. Thereafter, closure of the fistula was observed in all patients and this required from 10 to 25 days.

In the remaining 9 patients who presented after surgery with jaundice or cholangitis, ERCP detected cyst remnants in the biliary tract in all patients; also in 2 patients (22%) was observed associated choledocholithiasis: the toilette of VBP was achieved in all cases with Dormia basket. All patients underwent ES and removal of cyst remnants without complications (Table II).

Discussion

Our experience suggests that ES is safe and effective for management of biliary complications of hepatic hydatid disease. All patients remained asymptomatic after ES and there were no major complications from the endoscopic procedure: we registered 3 cases of transitory hyperamylasemia and one case of papillary bleeding after sphincterotomy, that was treated with blood transfusion.

The classic treatment for hydatid disease with rupture into the biliary tract has been surgery, with exploration of the biliary tract with choledochotomy, placement of a T-tube, and clearance of cyst remnants. In cases of papillary stenosis, additional manipulation is necessary in the form of sphincteroplasty or choledochoduodenostomy (6-8).

Nevertheless, the contribution of endoscopic clearance of biliary tract in the management of these patients has been significant, especially those with cholangitis. As treatment for acute cholangitis caused by choledocholithiasis, ES is superior to emergency surgery, the same may be expected for cholangitis caused by hydatid disease in non-surgically treated patients (9).

Cholestasis is a common complication of hepatic hydatid cysts rupture into the biliary tree, either spontaneously or following surgery, which occurs 4-20% of cases (10).

In literature, there are only six cases of ruptured echinococcus liver cyst into biliary tree which were completely treated by means ERCP and medical therapy (10-11): ERCP could be successfully employed to evacuate biliary daughter cysts and, eventually, to irrigate by naso-biliary drain hypertonic saline into the main cyst, resulting in complete cure of the disease.

The first and most common postoperative complication of surgery for hydatid cyst is persistent external drainage, in which there is a communication between the residual cyst and the biliary tree. Biliary fistula develops after surgical drainage of liver cysts in approximately 4% of patients (12). This occurs mainly because of the increased pressure in the biliary system. Surgery has been the traditional therapy for biliary fistulas because of hydatid cysts that fail to close spontaneously. However, because of the presence of adhesions and inflammation, surgery is difficult and hazardous (13-14). Endoscopic techniques have been developed to decrease the pressure in the biliary tract and reduce the time to closure of the fistula. Biliary ES, naso-biliary drainage, and endoprosthesis insertion have been used (15): the naso-biliary catheter decreases pressure in the biliary tract while allowing monitoring of the fistula by cholangiography. Its great advantage is that it is easily removed and that a cholangiogram can be obtained prior to removal.

However, these catheters are poorly tolerated, are subject to unexpected dislodgement, and probably prolong hospitalisation.

An endoprosthesis may be used subsequent to sphincterotomy; this is known to be effective in the treatment of postcholecystectomy fistulae, and the prosthesis may alternatively be placed without sphincterotomy, to be withdrawn when external drainage ceases (16-17).

In hydatidosis, ES is preferable because of the frequent occurrence of papillary stenosis or associated difficulties with drainage of bile and is also better tolerated by patients (18).

In literature is reported that naso-biliary drainage with continuous suction led to healing of a fistula that had not resolved after ES alone. Placement of the naso-biliary drain proximal to the defect allows continuous aspiration, and is especially effective when the fistula originates from the intrahepatic biliary tract (19).

In contrast to postcholecystectomy fistulae, which close rapidly after sphincterotomy and occasionally even do so spontaneously, hydatid fistulae are more resistant and long-lived and rarely close spontaneously without decompression of the biliary tract. Although the closure time for our series is admittedly longer than that reported for other types of fistula, we believe that in spite of this sphincterotomy is beneficial because it permits resolution and is a relatively conservative form of treatment (18). Data from several series indicate that the time frame for closure of external fistulas ranges from 3 to 43 days after ES (20-21). In our series, all 8 patients with persistent external drainage were treated successfully by ES and positioning of naso-biliary drainage, and the fistulas closed within 10 to 25 days.

It has also been suggested that preoperative endoscopic treatment may prevent the development of persistent external biliary drainage from the residual cavity (22). However, there are no published data to support the routine use of ERCP in the preoperative assessment of patients with hydatid disease without biliary complications; in these cases, we recur to MNRC or computed tomography (CT) (23-24).

The second common postoperative complication of hydatid cyst is jaundice and cholangitis caused by obstruction of the bile duct by hydatid remnants ES and clearance of the biliary tract is the treatment of choice (11). In the our series, all 9 patients with postoperative obstructive jaundice or cholangitis had cyst remnants in the bile duct, but two had also associated choledocholithiasis. With ES and clearance of the tract, symptoms resolved within 36 hours and all patients have remained asymptomatic during extended follow-up.

Conclusions

In our opinion, and according to the literature, ERCP + ES is safe and effective for the management of biliary complications of hepatic hydatid cysts, either before or after surgery, that is the only method to remove internally the cyst and prevent complications and relapses.

When is performed before surgery, make possible to clean out the biliary tree (hydatid remnants or daughter-cysts), the topographic evaluation and the biliary tree involvement (extrinsic compression).

With the positioning of naso-biliary drain, significantly assists in the diagnosis and, to a great degree, conclusively aids in the therapeutic task stemming from postoperative complications of biliary echinococcal disease, without the intrinsic risk of surgery; it should be considered the treatment of choice for postoperative biliary fistula.

The efficacy of preoperative endoscopic treatment for prevention of postoperative external leakage remain to be demonstrated and more studies of this problem should be carried out: we are in disagree with the role of preoperative ERCP, that is only operative and not diagnostic technique. That this endoscopic approach may achieve cure in such complicated cases should be kept in mind, but we underline that the only effective and curative therapy for hydatid liver disease is surgery.

Abstract

Background: The aim of this study was to evaluate the effectiveness of endoscopic sphincterotomy and positioning of naso-biliary drain or biliary endoprosthesis for preoperative and postoperative complications of hepatic hydatid disease (fistulas, compression cholestasis, rupture in biliary tree).

Methods: During the period 1994-2003, 22 patients (12 male and 10 female, mean age 55.4 years, range 16-65 yrs) underwent endoscopic treatment for complications of hepatic hydatid disease. Indications for ERCP in 5 patients treated before surgery (Group A) were obstructive jaundice in 4 (80%) and acute pancreatitis in 1 (20%). In 17 patients treated after surgery (Group B), the indication was acute cholangitis in 6 (36%), obstructive jaundice 3 (17%), and persistent external drainage in 8 patients (47%).

Observations: In group A, ERCP detected hydatid vesicles within the bile duct in 4 patients. 4 patients underwent endoscopic sphincterotomy and clearance of the duct with no complications. The 8 patients in Group B with persistent external drainage had biliary fistulas that resolved after endoscopic treatment within 10 to 25 days (in 4 patients with low-flow fistula was positioned nasobiliary drain to repeat cholangiogram) or in 4-6 weeks (in 4 patients with high-flow fistula was positioned biliary endoprosthesis). In 2 of 8 patients (25%) with postoperative external biliary fistulas was observed choledocholithiasis (resolution with ERCP). 4 the 9 patients with postoperative obstructive jaundice or acute cholangitis, had cyst remnants obstructing the bile

duct. Surgical reintervention was avoided in all patients in group B: all underwent endoscopic sphincterotomy and clearance of the bile duct without complications. After treatment, all patients remained asymptomatic.

Conclusion: Endoscopic sphincterotomy with positioning of naso-biliary drain or biliary endoprosthesis is a safe and effective treatment for preoperative and postoperative biliary complications of hepatic hydatid disease.

Key words: Hydatid disease, echinococcus, complication, management, ERCP.

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Commento

Commentary

Prof. Alberto LIBONI
Ordinario di Chirurgia Generale
Prof. Paolo ZAMBONI
Istituto di Chirurgia Generale
Università di Ferrara

Abbiamo lavorato per 5 anni presso l'Istituto di Patologia Chirurgica dell'Università di Sassari, in Sardegna, zona endemica per l'idatidosi. Nel corso di numerosi interventi di pericistectomia totale per echinococcosi epatica abbiamo osservato quasi costantemente una comunicazione fra la parete della cisti e le adiacenti vie biliari intraepatiche, anche in assenza di manifestazioni cliniche tipo ittero ostruttivo e/o colangiti acute biliosettiche. Questo rende ragione della elevata frequenza delle complicanze biliari ascrivibili alla cisti idatidea. La ERCP, il Dormia, la ES sono metodiche endoscopiche di indubbia utilità nella gestione delle complicanze pre e post operatorie di questa difficile affezione e, come dimostrato dal lavoro, vanno tenute presenti nella gestione globale della patologia. La pericistectomia totale e l'inserzione di un tubo di Kehr rimane comunque l'ottimale e definitivo trattamento chirurgico.

We spent 5 y in Sassari University, in Northern Sardinia, an endemic area for echinococcosis. We performed a large series of total pericystectomy owing such an epidemiology. Intra-operative inspection of the cystic wall constantly demonstrated at least one communication with the intra-hepatic biliary tree, even in absence of any clinical manifestation. This observation helps to understand the high frequency of the biliary complications observed by the Authors. In addition, they demonstrated that ERCP complemented by basket Dormia and endoscopic sphincterotomy is a useful tool for managing both preoperative and postoperative biliary complications of hepatic hydatid cyst. Such option has to be taken into account even if, only surgery allows to achieve the complete resolution of the disease by total pericystectomy and eventual T Tube insertion.

Autore corrispondente:

Ill.mo Prof. Carmelo SCIUMÈ
Via Eduardo Carapella, 12
00129 PALERMO