Acute pancreatitis during liver hydatidosis: treatment with ERCP and endoscopic sphincterotomy

INTRODUCTION: The Authors report on a case of a young woman who developed acute pancreatitis when affected by liver hydatidosis, successfully treated with endoscopic retrograde cholangiopancreatography (ERCP) and endoscopic sphincterotomy.

METHODS: An endoscopic sphincterotomy was performed, with extraction of multiple hydatid membranes.

OBSERVATIONS: Laboratory values returned to normal within 36 hours of the sphincterotomy. The patient was dismissed with oral therapy (Albendazole 400 mg bis in die for 4 months) and antibodies to Echinococcus were not detectable 1 month later. One year later, at ultrasound and CT the hydatid cyst was regressed and patient was still without symptoms.

CONCLUSIONS: Hydatid membranes in the biliary tract should be considered as a potential cause of pancreatitis in patients with hydatidosis, even if it is a rare complication, caused by the obstruction of the distal part of common bile duct by fragments of hydatid membranes, scolices or daughter cysts. ERCP may be resolutive, but surgery remains the treatment of choice for treatment of liver hydatid cysts.

KEY WORDS: Acute pancreatitis, Complications, ERCP, Liver hydatidosis (echinococcosis).

Hydatidosis is a zoonosis that is generally caused by infection with tapeworms (Echinococcus granulosus). When the parasite eggs are ingested by human beings, the infectious forms penetrate the intestinal wall and reach the liver via the portal circulation. In some cases, the hydatid cyst subsequently ruptures into the biliary tract.

Patients who have pancreatitis caused by hydatid membranes in the biliary tract have been treated, generally, by surgery.

We have encountered one case of acute pancreatitis caused by the presence of hydatid membranes in the biliary tract and in which the problem was resolved by endoscopic sphincterotomy.

Case report

A 18-year-old female was referred to our hospital with epigastric pain radiating to the back, jaundice, fever, nausea and emesis over a period of 12 hours. There was no history of drug or ethanol abuse. The patient had a history of hepatic hydatidosis (VII-VIII segments, diameter about 6 cm). Epigastric tenderness was found on physical examination. The most relevant laboratory results are summarized in Table I.

An abdominal US and CT confirmed a 6 cm diameter hydatid cyst in the right lobe with dilatation of biliary tract (12 mm) and irregular echogenic material in the lumen; the pancreas was enlarged and there were several peripancreatic fluid accumulations together with a little left pleural effusion.

ERCP (Fig. 1) revealed a normal main duodenal papilla with dilated common bile duct. On cholangiography, a dilated biliary tract (15 mm) containing multiple filling defects, mobile and with irregular morphology was seen. The pancreatogram was normal. An endoscopic sphincterotomy was performed and multiple hydatid membranes were extracted with a balloon catheter and Dormia basket (flat, green-yellowish membranes were extracted). The patient progressed satisfactorily, labora-
tory values returned to normal within 36 hours of the sphincterotomy. The patient was dismissed with oral therapy (Albendazole 400 mg bis in die for 4 months) and antibodies to Echinococcus were not detectable 1 month later. One year later, the hydatid cyst was regressed and patient was still without symptoms.

**Discussion**

Hepatic hydatidosis is a common disease in countries where the livestock are guarded by dogs \(^1\). It is endemic in Italy, with an annual incidence of 50-200 cases per 100000 inhabitants (Sardinia 14.32 cases per 100000, Sicily 4.5 casi / 100000) \(^2\). Spontaneous rupture of the hepatic hydatid cyst into the biliary tract occurs in 3.2% to 37% of the cases \(^{1,3}\), frank perforation in 3.2-17% and occult rupture in 10-37% \(^1\). Intrabiliary rupture of echinococcal cyst has been classified as either frank perforation with overt passage of hydatid material into the biliary tract or as occult leakage with signs of suppuration alone \(^1\) into three types: (a) contained rupture, referring to perforation of the true parasitic cystic wall within the host-derived pericyst, (b) communicating rupture, describing evacuation of cyst contents via anatomical diversion structures and (c) direct rupture (e.g. spilling of cyst material by a non-physiological route into preformed body cavities or adjacent hollow viscus) \(^1\). Rupture of the cyst into the biliary tract is associated with abdominal pain in 92% to 100% of the cases, with jaundice in 79% to 100%, fever in 60% to 93%, and cholangitis in 20% to 67% \(^1\). Less often, urticaria, anaphylactic shock, obstructive jaundice, secondary infection of the cyst, or dissemination to other

**Table I – Laboratory results**

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Admission</th>
<th>Post-ERCP</th>
<th>Dismission</th>
<th>Normal range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Amylase</td>
<td>1250</td>
<td>200</td>
<td>130</td>
<td>12-125 IU/L</td>
</tr>
<tr>
<td>Lipase</td>
<td>3220</td>
<td>1125</td>
<td>220</td>
<td>1-190 IU/L</td>
</tr>
<tr>
<td>Alanine aminotransferase</td>
<td>150</td>
<td>100</td>
<td>39</td>
<td>5-37 IU/L</td>
</tr>
<tr>
<td>Aspartate aminotransferase</td>
<td>230</td>
<td>122</td>
<td>53</td>
<td>5-40 IU/L</td>
</tr>
<tr>
<td>Gamma-Glutamyl transpeptidase</td>
<td>360</td>
<td>154</td>
<td>48</td>
<td>5-50 IU/L</td>
</tr>
<tr>
<td>Alkaline phosphatase</td>
<td>481</td>
<td>218</td>
<td>223</td>
<td>98-279 IU/L</td>
</tr>
<tr>
<td>Bilirubin</td>
<td>9.7</td>
<td>4.1</td>
<td>1.0</td>
<td>0.1-0.6 mg/dL</td>
</tr>
<tr>
<td>Leukocytes</td>
<td>18630</td>
<td>7300</td>
<td>5420</td>
<td>4.800-10.800/muL</td>
</tr>
</tbody>
</table>

Fig. 1: a) Dilated biliary tract (15 mm) containing multiple filling defects, mobile and with irregular morphology; b-c-d) Extraction of multiple hydatid membranes.
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In conclusion, one case of acute pancreatitis clearly related to rupture of hydatid cyst into the biliary tract have been presented. Hydatid membranes in the biliary tract should be considered as a potential cause of pancreatitis in patients with hydatidosis. This possibility should be taken into consideration in patients presenting with pancreatitis and hepatic hydatid cyst even if it is a rare complication, caused by the obstruction of the distal part of common bile duct by fragments of hydatid membranes, scolices or daughter cysts.

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Riassunto

INTRODUZIONE: Viene descritto il caso di una giovane donna portatrice di cisti epatica da echinococco che ha sviluppato un episodio di pancreatite acuta, trattata con successo mediante una ERCP con sfinterotomia endoscopica.

METODO: È stata eseguita una ERCP con sfinterotomia endoscopica (SE) ed estrazione di multiple caratteristiche membrane idatidee.

OSSERVAZIONI: I test di laboratorio sono andati incontro a normalizzazione a 36 ore dalla sfinterotomia. La paziente è stata quindi dimessa con terapia medica (Alben-dazolo 400 mg, 2 compresse al giorno per 4 mesi) e ad 1 mese il titolo anticroplasma è stato determinabile.

UN anno dopo, la cisti epatica non era più valutabile in imaging e la paziente è rimasta asintomatica.

CONCLUSIONE: Membrane idatidee nella via biliare possono essere considerate causa potenziale di pancreatite acuta da ostruzione. In tali casi, la ERCP può essere risolutive, ma la chirurgia rimane il trattamento di scelta per l'idatidosi epatica.

References

17) Alonso Casado O, Moreno Gonzalez E, Loinaz Segurola C,
Commento

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La segnalazione è incentrata su di un episodio di pancreatite da ostacolato deflusso del Wirsung per la presenza di membrane idatidee nella V.B.P., secondaria a rottura di una cisti epatica nelle Vie Biliari. La condizione è stata favorevolmente risolta mediante disostruzione attuata per via endoscopica (ERCP).

La segnalazione ha interesse anche per la coincidenza di numerosi fortunati eventi che hanno portato alla guarigione spontanea della cisti. La giovane età della paziente (18 anni) con fegato soffice, la verosimile giovane età della cisti a pericistio sottile, la rottura e completo svuotamento del materiale idatideo in un dotto biliare di calibro adeguato che è stato possibile rimuovere per via endoscopica, la mancata infezione e la spontanea elisione del cavo.

Qualora un ulteriore controllo a distanza, mediante TC spirale, dimostrasse la scomparsa della cisti, darebbe conferma della rara fortunata possibilità di una guarigione spontanea.

Ritengo, tuttavia, che di regola non si possa fare affidamento sulla coincidenza di tanti favorevoli fattori e, in presenza di una cisti di 6 cm, la condotta corretta comporti l’indicazione ad un intervento chirurgico possibilmente radicale (pericistectomia).

Only few cases of acute pancreatitis, as a complication of ruptured liver hydatid cyst, have been described. This case is about the recovery from a secondary acute pancreatitis caused by the rupture of a liver hydatid cyst into the biliary tract obstruction of the Wirsung by hydatid membranes. The pancreatitis was treated successfully by endoscopic removal of membrane fragments from the obstructed biliary tract.

It is important to highlight some concomitant events which led to spontaneous recovery of the cyst: the patient is a young woman (18 year old), with soft liver; the young hydatid cyst had thin pericyst; the burst of the cyst and the complete emptying of it into a biliary tract of suitable dimension.

Another favourable event was the spontaneous elision of the cyst cavity without infection. If the spiral CT, during future follow-up (2 years), showed the disappearance of the cyst, the rare lucky possibility of its spontaneous recovery would be confirmed.

However, from our experience, we cannot count on such favourable events for a 6 cm hydatid cyst. We would strongly recommend, whenever possible, as a correct therapeutic choice, radical surgery (pericystectomy).

Surgery remains the treatment of choice for the management of hydatid cyst.

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