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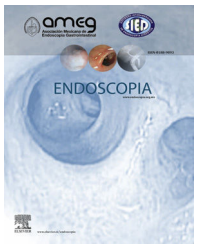

CASE SERIES

Subcapsular hepatic hematoma: An unusual, but potentially life-threating post-ERCP complication. Case report and literature review☆

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KEYWORDS
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Abstract Endoscopic retrograde cholangiopancreatography is a very common therapeutic procedure for a variety of biliary and pancreatic duct diseases. Procedure related complications are more frequent than in other endoscopic procedures. The most common reported complications are pancreatitis, bleeding, perforation, and infection. Subcapsular hepatic hematoma secondary to endoscopic retrograde cholangiopancreatography is a rare complication, but is potentially life-threating. The case is presented of a 25 year old patient with choledocholithiasis and cholelithiasis, who developed a subcapsular hepatic hematoma. The patient presented with

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an acute rupture of Glisson's capsule, which was successfully managed with surgical treatment. A review of the literature is presented, which consists of 30 cases.

PALABRAS CLAVE
Colangiopancreatografía retrógrada endoscópica retrograda;
Conducto biliar;
Hematoma;
Cirugía

Hematoma subcapsular hepático: una complicación post-CPRE poco frecuente pero potencialmente mortal. Reporte de caso y revisión de la literatura

Resumen La colangiopancreatografía retrógrada endoscópica es actualmente un procedimiento terapéutico muy común para el manejo de enfermedades biliopancreáticas. Las complicaciones secundarias a este procedimiento son más frecuentes que en otros procedimientos endoscópicos, dentro de las cuales la pancreatitis, hemorragia, perforación e infección son las que se presentan más a menudo. El hematoma hepático subcapsular secundario a colangiopancreatografía retrógrada endoscópica es una complicación muy rara, sin embargo esta puede poner en peligro la vida del paciente. Presentamos el caso de una paciente de 25 años con coledocolitiasis y colelitiasis que desarrolló un hematoma hepático subcapsular poscolangiopancreatografía retrógrada endoscópica con abdomen agudo y ruptura de la cápsula de Glisson que fue manejada exitosamente con tratamiento quirúrgico, así como una revisión de la literatura de 30 casos.

Introduction

Endoscopic retrograde cholangiopancreatography (ERCP) is widely available in most hospitals and is one of the most frequently performed procedures in the treatment of biliary–pancreatic diseases. Even in centers with a high-volume of patients that present with biliary and pancreatic pathology and are subsequently treated, serious complications from therapeutic ERCP can occur in 2.5–8% of cases, with mortality ranging from 0.5% to 1.0%.[1,2] Acute pancreatitis, cholangitis, hemorrhage, and perforation, the leading complications in 1–7%, 1.4%, 1%, and 1% of patients treated via ERCP respectively.[2,3] Subcapsular hepatic hematoma is a rare complication following ERCP and can be life-threatening in some cases.[4] The purpose of our study is to present a case of post-ERCP subcapsular hepatic hematoma which evolved with hypovolemic shock and required surgical intervention; we also review the possible pathophysiological mechanisms underlying such complication and analyze the sum of the contemporary available literature (Table 1).

Case report

A 25-year-old female with a history of pregnancy and C section 7 months prior to admission presented with recurrent abdominal pain in the upper right quadrant 15 days prior admission, the pain was accompanied by nausea, vomiting, and jaundice. Laboratory report showed the following: WBC $5.51 \times 1.000/\mu L$, Hemoglobin 11.5 g/dL, platellet count of $310 \times 1.000/\mu L$, prothrombin time ratio 13.3, international normalized ratio 1.16, aspartate aminotransferase 128 U/L, alanine aminotransferase 125 U/L, Total Bilirubin 3.57 mg/dL, Direct Bilirubin 2.96 mg/dL, alkaline phosphatase 928 U/L. An MRCP revealed intra and extrahepatic bile duct dilation with a defect in the distal portion of the common bile duct. ERCP with biliary sphincterotomy over a 0.035-inch diameter guidewire was performed, cannulation of the papilla on a first attempt without difficulty following the insertion of a 16 mm stone extraction balloon catheter (Shaili Endoscopy®) obtaining one yellow round bile duct stone of 5 mm of diameter (Fig. 1), control cholangiography showed no residual defects and the procedure

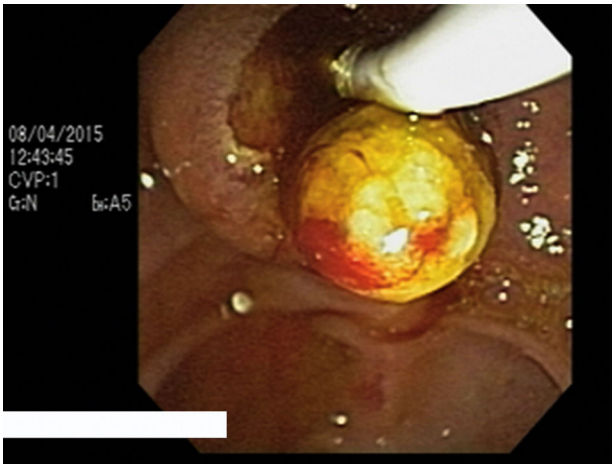

Figure 1 ERCP obtaining one yellow round bile duct stone of 5 mm of diameter.

Table 1 Hepatic subcapsular hematoma after ERCP summary of cases.

Reference	Age	Sex	Guidewire	Procedure	Comorbidity	Diagnostic Method	Hematoma rupture	Onset of symptoms
De la Serna et al.,[1,6,8]	71	F	Yes	Sphyncterotomy + stone extraction	None	CT	No	48 h
Horn et al.[1,6,8]	88	F	Yes	Biliary brushing + stent 10 Fr × 7 cm	Systemic hypertension + cholecystectomy	NA	No	48 h
Cardenas et al[1,6,8]	54	M	Yes	Sphyncterotomy + stent 10 Fr × 7 cm	Orthotopic liver transplant	CT	No	24 h
Gonzalez et al[2]	30	F	NA	Spyncterotomy + biliary ballon dilation + stent 10 Fr	None	NA	No	72 h
Oliveira et al[7]	84	M	NA	Stone extraction	COPD + pulmonary thromboembolism + anticoagulation	CT	No	10 days
Chi et al[1,6,8]	43	F	Yes	Sphyncterotomy + metalic stent + biliary balloon dilation 20 mm	Cholecystectomy + pancreatic cancer	CT	Yes	5 h
McArthur et al[1,6,8]	71	M	Yes	Sphyncterotomy + stone extraction + stent 7 Fr	None	CT	No	12 h
Baudet et al[1,6]	69	F	Yes	Sphyncterotomy + stone extraction	None	CT	Yes	48 h
Del Rosi et al[9]	28	F	Yes	Sphyncterotomy + stone extraction + stent 10 Fr × 10 cm	Cholecystectomy	CT	No	48 h
Carrica et al[4]	37	F	Yes	Sphyncterotomy + stone extraction	Cholecystectomy	MRI	No	72 h
Bartolo et al[3]	66	F	NA	Sphyncterotomy + stone extraction	None	NA	No	Immediate
Priego et al[1,6]	30	F	NA	Sphyncterotomy	None	CT	No	Immediate
Revuelto et al[1,6,8]	41	NA	NA	Sphyncterotomy + stone extraction	NA	CT	No	NA
Del Pozo et al[1,6]	76	F	Yes	Sphyncterotomy + stone extraction	Atrial fibrillation + anticoagulation	NA	No	6 h
Bhati et al[1,6,8]	51	F	Yes	Sphyncterotomy + stone extraction	None	CT	Yes	Immediate
Fei et al[1,6]	56	M	Yes	Sphyncterotomy + stone extraction	None	CT	No	72 h
Zizzo et al[1]	52	F	Yes	Sphyncterotomy + stone extraction	None	CT	No	24 h
Klímová et al[6]	52	M	Yes	Sphyncterotomy + stone extraction	Chronic pancreatitis	CT	No	6 h
Perez et al[1,6]	72	F	NA	Sphyncterotomy + stone extraction	Diabetes mellitus + systemic hypertension	NA	Yes	2 h
Orellana et al[1]	96	NA	NA	Ampullary biopsies + stent	Periampullary tumor	CT	No	4 h
Orellana et al[1,6]	49	M	NA	Biliary stent replacement	Testicular cancer + acute pancreatitis	CT	Yes	2 h
Orellana et al[1,6]	55	F	NA	Biliary stent replacement	Gallbladder cancer	CT	No	NA
Papachristou et al[8]	69	M	Yes	Billiary balloon dilation + brushing + ampullary biopsies + stent	Primary sclerosing cholangitis	CT	NA	48 h
De Mayo et al[8]	96	M	NA	Sphyncterotomy + stent	Acute pancreatitis	CT	No	4 h
Nari et al[1,6,8]	15	F	NA	NA	None	US	No	NA
Ortega et al[1,6,8]	81	M	NA	Sphyncterotomy + stone extraction	NA	NA	NA	NA
Bhandarkar et al[8]	64	F	NA	NA	NA	NA	NA	10 days
Ertugrul et al[1,6,8]	41	M	Yes	Stent	Colangiocarcinoma	CT	No	48 h
Petit et al[1,6,8]	98	M	Yes	Sphyncterotomy + stone extraction	Acute myocardial infarction + parcial gastrectomy sec. to peptic ulcer disease	CT	NA	48 h
Poon et al[10]	79	F	NA	Sphyncterotomy + stent	Stroke + congestive heart failure	CT	No	Immediate
Case report	25	F	Yes	Sphyncterotomy + stone extraction	None	US	Yes	12 h

Table 1 (*Continued*)

Reference	Age	Sex	Clinical manifestacions	Treatment	Antibiotics	Hemoglobin decrease (g/dL)	Death
De la Serna et al.,[1,6,8]	71	F	RUC pain	Conservative	Yes	NA	No
Horn et al.[1,6,8]	88	F	RUC pain + anemia	Conservative	Yes	NA	No
Cardenas et al[1,6,8]	54	M	Mesogastric abdominal pain	Conservative	Yes	3.2 g/dL	No
Gonzalez et al[2]	30	F	RUC pain	Surgery	Yes	NA	Yes
Oliveira et al[7]	84	M	RUC pain	Percutaneous drainage	Yes	3.3 g/dL	Yes
Chi et al[1,6,8]	43	F	RUC pain + anemia	Embolization	Yes	NA	No
McArthur et al[1,6,8]	71	M	RUC pain	Conservative	Yes	NA	No
Baudet et al[1,6]	69	F	RUC pain + fever	Embolization + surgery	Yes CF	7.3 g/dL	No
Del Rosi et al[9]	28	F	RUC pain	Conservative	Yes	7.3 g/dL	No
Carrica et al[4]	37	F	RUC pain	Percutaneous drainage	Yes CF	1.3 g/dL	No
Bartolo et al[3]	66	F	Hypotension + tachycardia	Surgery	No	10.3 g/dL	Yes
Priego et al[1,6]	30	F	RUC pain	Surgery	Yes	NA	No
Revuelto et al[1,6,8]	41	NA	Anemia	Conservative	Yes	NA	No
Del Pozo et al[1,6]	76	F	RUC pain	Conservative	Yes	NA	No
Bhati et al[1,6,8]	51	F	RUC pain + hypotension	Percutaneous drainage	NA	NA	No
Fei et al[1,6]	56	M	RUC pain	Percutaneous drainage	Yes	NA	No
Zizzo et al[1]	52	F	Epigastric pain	Embolization	Yes	3.3 g/dL	No
Klímová et al[6]	52	M	RUC pain	Embolization	Yes	6 g/dL	No
Perez et al[1,6]	72	F	RUC pain	Surgery	NA	NA	No
Orellana et al[1]	96	NA	Right omalgia	Conservative	Yes	NA	No
Orellana et al[1,6]	49	M	RUC pain	Embolization	NA	NA	No
Orellana et al[1,6]	55	F	RUC pain + right omalgia	Conservative	NA	NA	No
Papachristou et al[8]	69	M	RUC pain + right omalgia + anemia	Conservative	NA	3.5 g/dL	NA
De Mayo et al[8]	96	M	Right omalgia	Conservative	Yes	NA	No
Nari et al[1,6,8]	15	F	RUC pain + right omalgia + hypotension	Conservative	Yes	NA	No
Ortega et al[1,6,8]	81	M	RUC pain	Percutaneous drainage	Yes	NA	NA
Bhandarkar et al[8]	64	F	RUC pain + fever + anemia	Percutaneous drainage	NA	NA	NA
Ertugrul et al[1,6,8]	41	M	RUC pain + fever	Conservative	Yes	NA	No
Petit et al[1,6,8]	98	M	RUC pain + asthenia	Percutaneous drainage	NA	NA	NA
Poon et al[10]	79	F	RUC pain + hypotension	Conservative	NA	5 g/dL	No
Case report	25	F	RUC pain + anemia	Surgery	Yes	4.2 g/dL	No

F – female, M – male, COPD – Chronic Obstructive Pulmonary Disease, RUC – right upper cuadrant, CT – computed tomography, US – ultrasound, MRI – magnetic resonance image, NA – not available, CF – *Citrobacter freundii*.

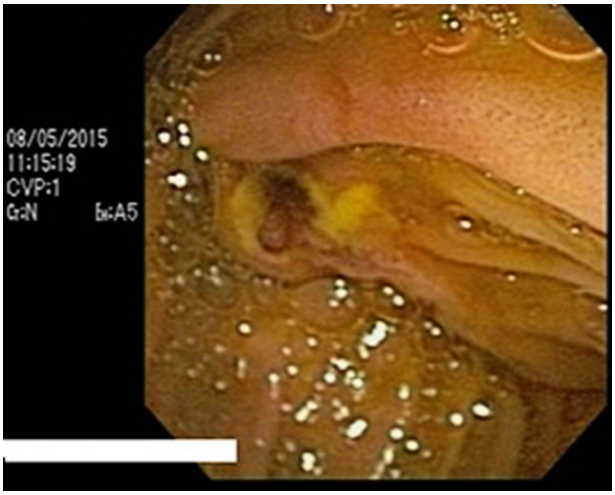

Figure 2 Upper endoscopy with duodenoscope without evidence of bleeding from sphyncterotomy.

was finished without evident complications such as bleeding or injury. 12 h post procedure the patient developed right upper quadrant abdominal pain, hemoglobin dropped 4 g/dL (Hb 7.3 g/dL), amylase spiked to 162 U/L and lipase also showed a rise to 213 U/L. There was no clinical evidence of gastrointestinal bleeding. An upper endoscopy was performed with a frontal video endoscope and a duodenoscope without evidence of gastrointestinal bleeding or post-sphyncterotomy bleeding (Fig. 2). Three packed red blood cells (PRBCs) were transfused, and a 12 h post transfusional hemoglobin was obtained which reported 10.3 g/dL. Hemoglobin declined to 5.3 g/dL 3 days after ERCP. The patient developed grade 4 hypovolemic shock with acute abdomen and positive Blumberg sign. An urgent abdominal ultrasound revealed free fluid in the abdominal cavity. Two more PRBCs were transfused and an urgent laparotomy was performed observing rupture of Glisson's capsule (Fig. 3) and approximately 3000 ml of blood in the peritoneal cavity was found. Abdominal packing was performed and two more PRBCs were transfused during surgery. The patient was admitted to the Intensive Care Unit (ICU) where she had to have assisted breathing via endotracheal tube. There was no need for vasoactive support. 48 h later a second laparotomy was performed to unpack the abdomen. The patient was extubated 6 days after the first laparotomy and was discharged from the ICU. The patient evolved well, maintaining hemoglobin levels at 10.9 g/dL and was discharged from the hospital 16 days after the ERCP procedure. One month later a follow-up MRI demonstrated a remanent heterogenic subcapsular hepatic hematoma of $15.2 \times 10.4 \times 3.6$ cm in the right hepatic lobe (Fig. 4). Currently, the patient is (3 months post ERCP) generally well and reports slight abdominal discomfort, patient surveillance will continue until complete resolution of the hematoma is observed.

Discussion

Bleeding related to an ERCP procedure is usually due to endoscopic sphincterotomy. Bleeding seen endoscopically during or immediately after sphincterotomy is not

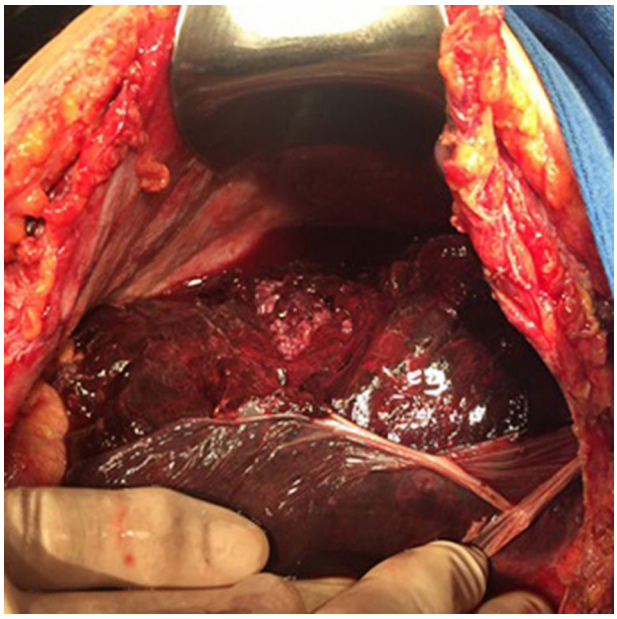

Figure 3 Rupture of Glisson's capsule observed in laparotomy.

Figure 4 Subcapsular hepatic hematoma of $15.2 \times 10.4 \times 3.6$ cm in the right hepatic lobe in magnetic resonance imaging (MRI) one month after presentationT2W sequence on a 1.5-T MRI machine.

uncommon, but is generally not considered an adverse event unless there is significant blood loss, which is manifested as evident hypovolemia and/or that has a need for transfusion. When applying clinical criteria such as melena, hematemesis, a drop greater than 2 g/dL in hemoglobin level, or requirement for secondary intervention such as endoscopy or blood transfusion, the overall incidence of bleeding is around 0.1–2% even in experienced hands.[5] Subcapsular hepatic hematoma is a rare and exceptional event, of which only 30 cases have been reported since 2000. The pathophysiologic mechanism underlying the development of the

hematoma is not clearly defined, however various authors agree that vascular injury of small-caliber intrahepatic vessels secondary to guidewire use may be the cause, which may also explain the presence of free air and development of infections in some of the cases.[1,6]

Patient factors

We found that the median age of presentation is 59 (15–98) years, with 58% female and 35% male distribution. Only 2 (6.4%) patients had a history of anticoagulation therapy prior to the procedure. Almost half of the patients (48.3%) presented clinical manifestations in the first 24 h post-ERCP. The most common manifestation was abdominal pain in 87%; in some case reports fever was mentioned to be one of the primary manifestations, but we found that right omalgia and clinical manifestations of hypovolemic shock (tachycardia or hypotension) were present more often 9.6% vs 16.1% vs 12.9%.

Procedure factor

In 17 (54.8%) cases the use of a guide wire was reported, but we cannot exclude its use on the other 14 cases. The literature on these 14 cases does not mention the use of a guide wire.

In most of the cases, 67.7%, diagnosis was performed by CT, but the use of US and MRI was also described in 2 and 1 cases respectively. In our case report abdominal US was performed searching for free fluid in the abdominal cavity, because of the development of an acute abdomen. In almost half of the patients (45.1%) no invasive treatment was required. Percutaneous drainage was performed in 7 (22.5%), surgery in 6 (19.3%), and embolization in 4 (12.9%) patients. Only 2 case reports described the pathogen found in the hematoma culture, both found *Citrobacter freundii*, which is a facultative aerobic gram-negative bacilli of the Enterobacteriaceae family, commonly found in the gastrointestinal tract. In 22 (70.9%) patients antibiotics where used, either as a prophylactic measure or as treatment for infection, 8 (25.8%) did not specify whether they used antibiotics or not. Only 11 case reports mention a decrease in hemoglobin that went between 1.3 g/dL to 10.3 g/dL, and from those cases, the majority, 63%, required some kind of intervention (embolization, percutaneous drainage or surgery), concluding that hemoglobin decrease is a good predictor for requiring intervention, no association was found between hemoglobin drop and the need of a specific kind of intervention. 3 (9.6%) deaths were reported, 2 patients who required surgery and one in whom percutaneous drainage was performed.

Because of the mortality rate found in the literature it is important to consider this as a life-threatening adverse event. Another rare complication following ERCP that can have a similar presentation as described in the case report is injury to the spleen causing intraperitoneal hemorrhage with a few cases being reported in the literature. Symptoms often include localized peritoneal irritation in the left upper quadrant progressing with time to generalized peritonitis, along with referred pain to the left shoulder. Changes in vital signs including tachycardia and orthostatic changes in blood pressure are frequent.

Conclusions

Subcapsular hepatic hematoma is a rare condition associated to ERCP that must be suspected when abdominal pain is present after the procedure, it may have a fatal course if it is not diagnosed and treated early. Further publications of case reports will help establish risk factors and appropriate treatment.

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Conflict of interests

Authors declare no conflict of interests for this article.

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