

Hemobilia Caused by Arterioportal Fistula Following Percutaneous Liver Biopsy Complicated by Acute Cholecystitis and Review of the Literature

ORESTIS IOANNIDIS¹, ANASTASIA MALLIORA¹, PANAGIOTIS CHRISTIDIS¹,
MANOUSOS GEORGE PRAMATEFTAKIS¹, EFSTATHIOS KOTIDIS¹,
IOANNIS MANTZOROS¹, NIKOLAOS OUZOUNIDIS¹, VASILIS FOUTSITZIS¹,
STAMATIOS ANGELOPOULOS¹

¹4th Academic Department of Surgery, School of Medicine,
Aristotle University of Thessaloniki, Greece

ABSTRACT: We present a case of a 44-year-old male with chronic hepatitis B that visited the Emergency Department due to epigastric pain after a liver biopsy. The ultrasonography revealed signs of bleeding in the bile ducts. and angiography visualized an arterioportal fistula. Selective right hepatic artery branch embolization was performed, and the bleeding was controlled. Although, the clinical picture was initially improved, the patient presented later with acute abdomen, obstructive jaundice and fever. The patient underwent cholecystectomy with bile duct exploration and placement of a Kehr's T tube in the common bile duct. The postoperative course was uneventful. We also review the relevant literature concerning arterioportal fistula manifested as hemobilia as well as acute cholecystitis occurring after hemobilia.

KEYWORDS: *Gastrointestinal bleeding, hepatic artery, hepatitis, jaundice, portal vein, hemobilia.*

Introduction

Hemobilia, a pathological communication between the bile ducts and the hepatic vascular circulation, is an unusual (0,006%) complication of percutaneous liver biopsy (PLB) [1], but it can be fatal [2].

Its clinical presentation is characterized by the triad of: upper gastrointestinal (GI) hemorrhage (90% of patients), right upper quadrant (RUQ) pain (70%) and jaundice (60%) [3].

Arterioportal fistula is an extremely rare complication of PLB presenting in about 5% of cases and usually remains asymptomatic while it is an even rarer source of hemobilia [4], that has been reported 12 times [3-14], to the best of our knowledge.

Acute cholecystitis is a very rare complication of hemobilia.

We describe a single case of hemobilia due to an arterioportal fistula presenting after a percutaneous liver biopsy that was treated with selective right hepatic artery branch embolization.

We also review the relevant literature concerning arterioportal fistula manifested as hemobilia after PLB as well as acute cholecystitis occurring after hemobilia.

A written informed consent was obtained from the patient for this publication.

Case Report

A 44-year-old male positive for HBsAg, HBV-DNA, anti-HBe with RUQ pain and elevated liver transaminase levels underwent a PLB without immediate complication.

The histological analysis of the liver biopsy revealed chronic hepatitis B lesions, without significant fibrosis.

On the 2nd post-biopsy day, he came to the Emergency Department (ED) with severe epigastric pain and jaundice.

His vital signs were within the normal limits.

Laboratory tests indicated elevated liver transaminase levels (SGOT=350mg/dL, SGPT=420mg/dL), hyperbilirubinemia with direct bilirubin predominance [total bilirubin (tBLR)=7,2mg/dL, direct bilirubin (dBLR)=4,7mg/dL] and elevated alkaline phosphatase (ALP) and gamma-glutamyl transferase (GGT), while his white blood count was normal.

He was admitted to the hospital and underwent an abdominal ultrasound, which was suspicious for a gallbladder hematoma (Figure 1).

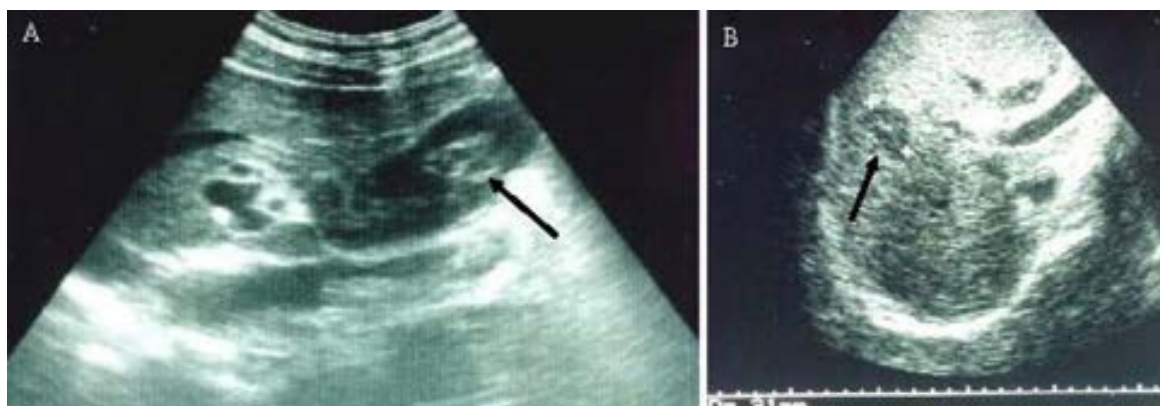


Figure 1. A. Ultrasound on the 2nd post-biopsy day demonstrating suspicion of gallbladder hematoma (arrow). B. Ultrasound showing a hypoechoic lesion of the right liver lobe (3cm) possibly due to focal ischemia following arterial embolization of a branch of the right hepatic artery (arrow).

On the 4th post-biopsy day, the RUQ pain and the jaundice were exacerbated (tBLR=10mg/dL, dBLR=5,6mg/dL), melena had developed, hematocrit dropped (Hct=35%) and RUQ ultrasound revealed gallbladder enlargement and heterogeneous content without acoustic shadow. The diagnosis of hemobilia after PLB was established.

The following days (5th-8th post-biopsy days), the clinical appearance was aggravated. Abdominal pain and melena increased, hematocrit continued to decrease (Hct=23,6%)

and transaminase levels increased (SGOT=1200mg/dL, SGPT 1300mg/dL).

The patient was transferred to the angiography unit for digital subtractive angiography for diagnosis and possible management of the hemobilia.

Angiography revealed a pathological communication between RUQ a branch of the right hepatic artery with a portal vein branch which was treated with selective embolization of the hepatic artery branch (Figure 2).

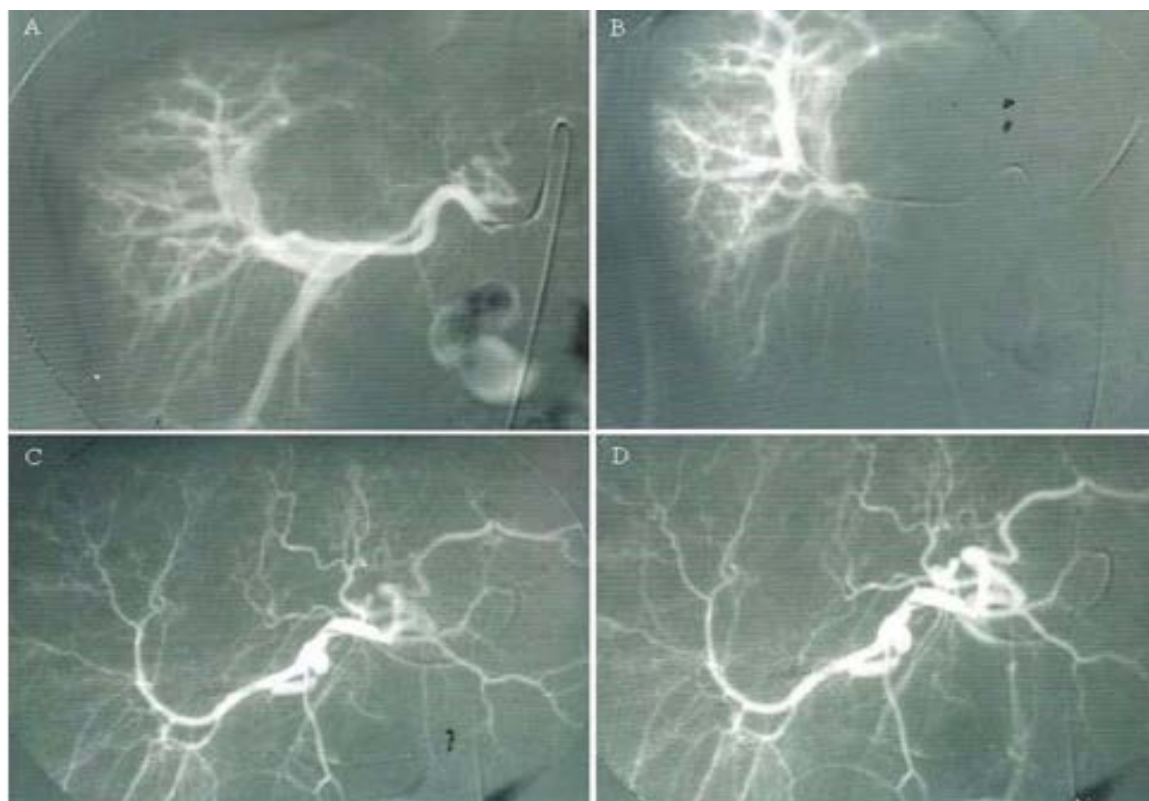


Figure 2. A. Angiography revealing pathological communication of a right hepatic artery branch with a right portal vein branch. B. Right hepatic artery branch embolization. C, D. Post-embolization angiogram.

The first days after the embolization (9th-10th post-biopsy/1st-2nd post-embolism days) the patient was stabilized hemodynamically and improved overall.

Ultrasound of the abdomen showed a hypoechoic lesion of the right liver lobe and an anechoic gallbladder (Figure 1).

However, elevated liver transaminase levels persisted.

The next week (12th-20th post-biopsy/3rd-11th post-embolism days), the patient's clinical status and laboratory tests meliorated.

Hematocrit increased (Ht=35,5%) and transaminase and BLR levels decreased (SGOT=49mg/dL, SGPT=102mg/dL, tBLR=1,3mg/dl) and the patient was discharged home after 18 days of hospitalization.

On the 31th post-biopsy/21th post-embolism day, the patient presented again to the ED due to fever, jaundice and abdominal pain.

His laboratory test revealed elevated white blood cell (WBC 14,800/mm³ with neutrophils 85%), hematocrit was 34%, SGOT 180mg/dL, SGPT 150mg/dL, tBLR 8,4mg/dL (dBLR 5,4mg/dL) and GGT 386mg/dL.

His clinical examination was indicative of acute abdomen with guarding and rebound tenderness especially at the RUQ.

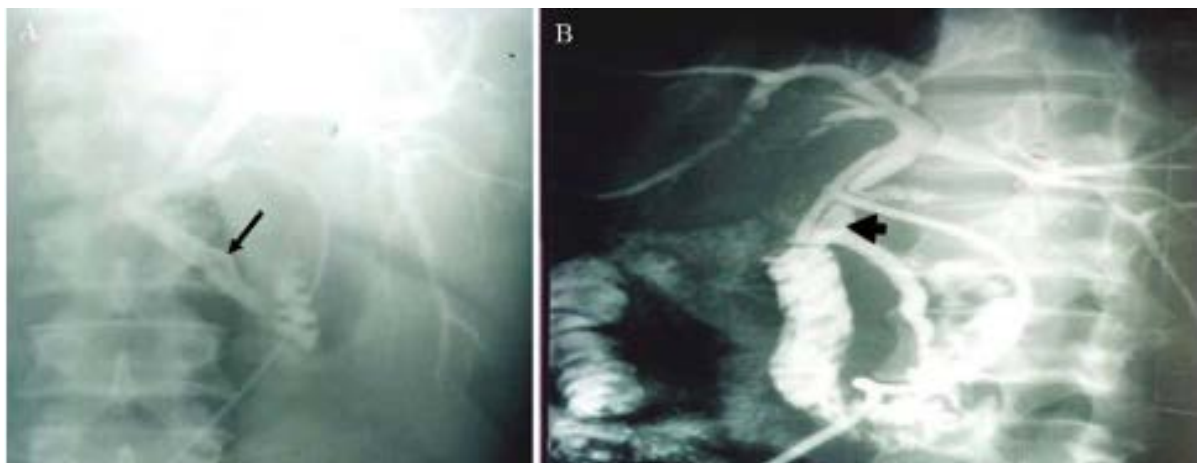
Ultrasound revealed gallbladder distention with wall thickening and hyperechoic content inside it, as well as cystic and common bile duct dilation, confirming the diagnosis of acute cholecystitis.

Thus, the patient underwent open surgery, in which he was submitted to cholecystectomy, intraoperative cholangiography which showed filling defects of the common bile duct (Figure 3), choledochotomy, common bile duct exploration, hemorrhagic clots removal and placement of a Kehr's T-tube in the common bile duct.

His post-operative course (2nd-7th postoperative days) was uneventful with improvement of his clinical status and laboratory tests.

The bile culture was negative, the T-tube cholangiography performed on the 7th post-operative day showed normal drainage of contrast medium to the duodenum, hepatic and common bile duct dilatation, and multiple small filling defects (possibly blood clots) (Figure 3).

On the 12th post-operative/43rd post-biopsy day, his clinical picture and laboratory tests improved further (Hct=37,8%, SGOT=74mg/dL, SGPT=102mg/dL, GGT=127mg/dL, tBLR=1,2mg/dL, dBLR=0,7mg/dL), T-tube was removed and the patient was discharged home.



**Figure 3. A. Intraoperative cholangiography: filling defects of the common bile duct (arrow).
B. T tube cholangiography showing small filling defects of the common bile duct (arrow).**

Discussion

Percutaneous liver biopsy is a frequent procedure that facilitates diagnosis and treatment of both acute and chronic liver diseases.

It is safe [1], but as every other procedure, it may lead to some complications.

It has been proved that complications manifest in 4,1% of the transthoracic approach and in 2,7% of the subcostal approach [3].

The complication rate seems to depend on the liver disease, the possible coagulopathy, the

type and the diameter of biopsy needles that are used and the number of needle insertion attempts [4] as well as the technique and the doctor's experience [7].

Bleeding is the most serious complication and it can present as free intraperitoneal hemorrhaging, hematoma or hemobilia [3].

Hemobilia, a pathological communication between the bile ducts and the hepatic vascular circulation, is an unusual (0,006%) complication of percutaneous liver biopsy [1], but it can be fatal [2].

In the past, the main cause of hemobilia was accidental injury of the abdomen, while nowadays its cause is iatrogenic in 17-58% of the cases owing to the development of new invasive, diagnostic and therapeutic methods for the hepatic-biliary system [3,4].

Its clinical presentation is characterized by the triad of: upper GI hemorrhage (90% of patients), RUQ pain (70%) and jaundice (60%)

[3] and it manifests usually within 5 days after the PLB [5].

In our case the last two symptoms presented on the 2nd post-biopsy day, while the first one on the 4th day.

Although ultrasonography and CT scan may play an important role on hemobilia diagnosis by revealing echogenic shadows due to clots in the bile ducts [15], angiography can show the source of the bleeding in the majority of cases [16] including ours.

More specifically, an angiogram can visualize an arteriobiliary fistula, a pseudoaneurysm or an arteriportal fistula.

In particular, arteriportal fistula is a rare (5%) complication of PLB and usually asymptomatic [4], in contrast to our case.

Hemobilia due to arteriportal fistula is extremely rare.

In the published literature, there are 12 cases of arteriportal fistula causing hemobilia after a PLB [3-14].

Table 1. Reported hemobilia cases due to arteriportal fistula after liver biopsy.

Case	Age	Gender	PLB cause	U/S-guided PLB	RUQ pain	Upper GI hemorrhage	Jaundice	Symptoms onset after PLB	Diagnostic tool	Treatment	Efficient treatment
Machicao et al. 2002 [3]	49	male	HCV	+	+	+	+	3 days	angiogram	embolization	+
Lin et al. 2005 [4]	68	female	HCV	+	+	-	-	2 days	angiogram	embolization	+
Zhou et al. 2014 [5]	57	female	abnormal liver function	+	+	+(melena)	+	7 days	angiogram	embolization	+
Gómez-Valero et al. 2001 [6]	67	female	chronic biochemical picture of cholestasis	+	+	+	+	2 days	angiogram	embolization	1st: - 2nd: +
Zhang et al. 2017 [7]	59	male	HCV	NM	+	+	+	6 days	angiogram	embolization	+
Hodgson et al. 2004 [8]	32	male	abnormal liver function	NM	+	+(hematochezia)	+	4 days	angiogram	embolization	+
Grieco et al. 1996 [9]	65	female	differential diagnosis of a non-homogenous hepatic structure revealed by USD	+	+	+(massive hematemesis and melena)	+	18-24 hours	ERCP, angiogram	embolization	+
Ormann et al. 1991 [12]	42	male	alcoholic liver cirrhosis	NM		severe hemobilia		5 days	angiogram	embolization	+
Labayle et al. 1978 [11]	62	male	abnormal liver function	Intraoperative	-	+	+	NM	angiogram	embolization	-

RUQ: right upper quadrant; u/s: ultrasound; PLB: percutaneous liver biopsy; GI: gastrointestinal; HCV: hepatitis C virus; ERCP: endoscopic retrograde cholangio-pancreatography; NM: not mentioned.

We managed to find 9 of the 12 full case reports which are summarized in Table 1.

The patients' mean age was found to be 55,6 years and the male/female ratio 5:4.

Symptoms onset was 3,75 days after PLB on average and the characteristic triad of hemobilia was manifested in 6 out of 9 patients.

More specifically, either RUQ pain or upper GI hemorrhage or jaundice occurred in 87,5%.

In all cases, embolization was chosen for the treatment of hemobilia.

In seven patients, this was efficient, while in one of them, the arterioportal fistula recanalized, so further embolization was required, which was able to control the hemobilia ultimately [3-14].

Hemobilia can be rarely complicated with acute cholecystitis.

To our knowledge, 14 cases have been reported [7,15,17-26], which are summarized in Table 2.

Table 2. Reported acute cholecystitis cases due to hemobilia.

Case	Age	Gender	Cause	Source	Hemobilia diagnostic tool	Acute cholecystitis diagnostic tool	Diagnosis of acute cholecystitis after hemobilia	Diagnosis of acute cholecystitis after PLB	Treatment
Zhou et al. 2014 [5]	57	female	PLB	arteriovenous fistula	angiogram	u/s	3 days after the onset of symptoms of hemobilia	12 days	percutaneous cholecystostomy
Egritas et al. 2010 [15]	7	female	PLB	NM	CT scan	u/s	simultaneously with the onset of symptoms of hemobilia	3 days	conservative
Albuquerque et al. 2005 [26]	21	female	PLB	NM	MRCP, ERCP	u/s	6 days after the onset of symptoms of hemobilia	7 days	cholecystectomy
Moon et al. 2006[17]	59	female	PLB	NM	NM	u/s	NM	2 days	NM
Priya et al. 2013 [20]	22	male	spontaneous	cystic artery gallbladder fistula	angiogram	u/s	simultaneously with hemobilia diagnosis	NM	cholecystectomy
Lee et al. 1999 [23]	30	female	PLB	NM	ERCP	u/s	simultaneously with the onset of symptoms of hemobilia	5 days	cholecystectomy
Edden et al. 2006 [24]	15	male	PLB	NM	u/s (a large blood clot in the gall bladder)	u/s	NM	12 days	cholecystectomy
Coelho et al. 2001 [25]	39	female	PLB	NM	intraoperative	u/s	NM	2 days	cholecystectomy
Counihan et al 1996 [18]	22	female	liver biopsy	NM	NM	NM	NM	NM	NM
Sumida et al. 2005 [19]	NM	NM	PLB	NM	NM	NM	NM	NM	NM
Maeda et al. 2002 [21]	62	male	NM	cystic artery pseudoaneurysm	angiogram	u/s	NM	NM	cholecystectomy
Lewis et al. 1982 [22]	29	male	PLB	arterioportal fistula	angiogram	u/s	simultaneously with the onset of symptoms of hemobilia	5 days	conservative - antibiotics

RUQ: right upper quadrant; u/s: ultrasound; PLB: percutaneous liver biopsy; GI: gastrointestinal; HCV: hepatitis C virus; ERCP: endoscopic retrograde cholangio-pancreatography; MRCP: Magnetic resonance cholangiopancreatography; CT: computed tomography; NM: not mentioned

The patients' average age was 39,6 years, whereas 53,85% of them were female.

In the majority of the cases (64,3%) hemobilia occurred secondary to PLB on the 6th post-biopsy day on average, while in about one third of the cases acute cholecystitis manifested simultaneously with the onset of symptoms of hemobilia.

The majority of patients with acute cholecystitis were treated with cholecystectomy (66,6%), while percutaneous cholecystostomy or conservative treatment was also an option.

Moreover, acute cholecystitis is a known complication of right hepatic artery embolization [27-29].

The cystic artery, a terminal branch, originates typically from the right hepatic artery (79,02%) [30] and supplies oxygenated blood to the gallbladder.

The terminal nature of blood supply of the gallbladder makes it vulnerable to ischemia [31].

Thus, in our case, in addition to hemobilia, the right hepatic artery branch embolization may have contributed to acute cholecystitis.

Hemobilia can be treated with surgical procedure or arterial embolization.

More specifically, arterial embolization, first used for this purpose in 1976 [32], has been proved to be a secure and efficient (90-95%) treatment of hemobilia with low morbidity [4,33].

The materials that are usually preferred for embolization are Gelfoam pledgets, steel coils, and PVA particles.

It is essential that the blood flow of portal vein is verified, because portal vein thrombosis makes arterial embolization hazardous [34].

If arterial embolization fails to control the hemobilia, laparotomy for hepatic arterial ligation or hepatectomy could be performed [4].

In our case the hemobilia was efficiently controlled with selective right hepatic artery branch embolization.

In conclusion, hemobilia is an unusual but possibly fatal complication of PLB, caused by an otherwise safe procedure.

Thus, in case of upper GI hemorrhage after a liver procedure, hemobilia should be considered and if verified, it should be treated as soon as possible.

The first therapeutic option should be angiography, which is capable of revealing the source of bleeding and simultaneously treating the cause of bleeding by embolizing.

In case that arterial embolization fails to control the hemobilia, surgery could be performed.

Acknowledgments

The authors have no one to acknowledge.

Conflict of interests

None to declare.

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Corresponding Author: Orestis Ioannidis, 4th Academic Department of Surgery, School of Medicine, Aristotle University of Thessaloniki, Alexandrou Mihailidi 13, 54640 Thessaloniki, Greece, e-mail: iorestis@auth.gr