Management of massive haemobilia in an Indian hospital

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Abstract

Introduction Massive haemobilia carries a mortality of 25% in most reports. Although previously it was mainly due to road accidents or homicidal attempts it is now more often due to iatrogenic trauma like percutaneous liver biopsy and biliary drainage. However the management protocol is not established and there have been few reports of this serious condition from India.

Aim To review the causes of massive haemobilia and outline its management in an Indian hospital.

Patients and methods We retrospectively analysed the records of 20 consecutive patients with massive haemobilia (blood requirement more than 1400 ml/day) admitted to our department over six years from a prospectively maintained database. There were 10 males and 10 females who had a mean age of 43 (range 15–65) years.

Results Haemobilia accounted for 9 percent of patients admitted with upper gastrointestinal bleeding who were seen over this period. The commonest cause was iatrogenic (11) including laparoscopic cholecystectomy (6), Whipple's operation, endoscopic retrograde cholangiography (ERC), percutaneous transhepatic cholangiography (PTC), hepatic stone extraction and removal of biliary stent (1 each). The others had accidental trauma (4), visceral aneurysms (2), biliary stones (2) and chronic pancreatitis (1). The commonest clinical presentation was massive gastrointestinal

bleeding. The dual phase computed tomography (CT) scan correctly identified the site of bleeding and other associated conditions in all the 11 patients in whom it was done. Conventional angiography was done in 8 patients with transarterial embolisation (TAE) being attempted in 6 and successful in 2 patients. Operations were performed in 18 patients for the following indications – failure of angiographic embolisation (6), failure of endoscopic sclerotherapy (EST) (1), duodenal erosion (2), portal biliopathy (1), haemoperitoneum (1), bile leak (1), pseudocyst (1), liver necrosis (1) and other hepatobiliary conditions (4). The surgical procedures to control bleeding were ligation of aneurysms (8), repair of the hepatic artery (4), right hepatectomy (3), lienorenal shunt, cholecystectomy and under-running of the duodenal papilla (1 each). The overall mortality was 4 patients (20 percent). There was no mortality in patients with bleeding aneurysms; the mortality being significantly higher in patients with non-aneurysmal bleeding ($p=0.0049$; Fishers' exact test).

Conclusions In our experience haemobilia was usually due to an iatrogenic cause with a pseudoaneurysm following a diagnostic or therapeutic intervention (most often laparoscopic cholecystectomy) being the commonest aetiology. A dual phase CT scan accurately identified the site of bleeding. Angiographic embolisation often failed to stop bleeding and mortality was significantly higher in patients with non-aneurysmal bleeding. We should perhaps consider early surgery for haemobilia once the bleeding site has been localised by CT scan.

Keywords Haemobilia · GI bleed · Liver trauma · Visceral aneurysm

Introduction

Haemobilia, though rare, may be life threatening. Its aetiology is varied and its exact incidence is unknown. This is because it is often minor, self-limiting and hence unreported.

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Furthermore massive haemobilia is difficult to diagnose due to its non-specific clinical features and a low index of suspicion. The classical triad of bleeding, jaundice and upper abdominal pain is present in only 22 percent of cases [1].

Over the last few years the aetiology of haemobilia has shown a shift from being due to accidents to now being due to iatrogenic trauma from diagnostic and therapeutic interventions. A review of the literature between 1996 to 1999 by Green et al [1] in 2001, reported an iatrogenic etiology in 66 percent of cases. Due to its rarity and difficult diagnosis an optimal management protocol remains unclear. Since its introduction in 1976, the trans-arterial embolisation (TAE) has gained repute as a first line intervention to stop bleeding [2–9]. Other interventional approaches like endoscopic sclerotherapy [10], embolisation of the needle liver biopsy tract [11] have also been reported as case snippets. These reports have encouraged the nonoperative, sophisticated, interventional modalities to be the first line of treatment. The role of surgery has been infrequently reported in the recent literature. In India where radiological intervention techniques and expertise are either not widely available or expensive, surgery is often the only option available to manage these patients.

Because of the paucity of reports from this country we review our experience with massive haemobilia in an Indian institution.

Aim

To identify the aetiology of massive haemobilia and outline its management protocol in an Indian hospital.

Methods

Out of 220 patients with upper gastrointestinal bleeding who underwent surgery, in the Department of Surgical Gas-

troenterology and Liver Transplantation, Sir Ganga Ram Hospital, New Delhi, 20 patients with massive haemobilia were evaluated. Haemobilia was defined as massive if it caused hypotension or if there was a blood requirement of more than four units in 24 hours to maintain haemodynamic stability. The diagnosis was made after demonstration of the site of bleeding by upper gastrointestinal endoscopy, CT scan, angiography or surgery. Angiography whenever performed was followed by an attempt of TAE. Surgery was performed to control bleeding and to treat underlying associated conditions.

Results

There were 20 patients, 10 males and 10 females, who had a mean age of 43 (range 15–65) years.

Aetiology

The commonest aetiology was iatrogenic trauma which accounted for 11 (55 percent) patients (Table 1). These included 6 patients who had had a recent laparoscopic cholecystectomy (LC) and 1 each following Whipple's operation, hepatic stone extraction with U-tube placement, endoscopic retrograde cholangiography (ERC), removal of biliary stent and percutaneous transhepatic cholangiography (PTC).

Spontaneous bleeding, without external intervention, occurred in 5 patients. A retained stone in the common hepatic duct following cholecystectomy and choledochoduodenostomy was present in 2 patients. The stones had eroded into the right and left hepatic arteries respectively leading to formation of pseudoaneurysms in these patients. Two patients had visceral aneurysms (one of the gastroduodenal artery and the other had multiple intrahepatic aneurysms). Chronic pancreatitis with a pseudocyst, portal cavernoma and a pseudoaneurysm of the gastroduodenal artery

Table 1 Aetiology of iatrogenic massive haemobilia

Iatrogenic trauma (n=11, 55 percent)			
Initial diagnosis	Procedure	Cause of bleeding	n
Cholelithiasis	LC	Pseudoaneurysm RHA	3
		Pseudoaneurysm anterior branch of RHA	1
		Pseudoaneurysm RHA with tear in CHD	1
		Tear in RHA and CHD	1
Ampullary carcinoma	Whipple's procedure	Pseudoaneurysm GDA eroding into HJ	1
Hepatolithiasis	HJ and U-tube placement	Pseudoaneurysm anterior branch of RHA	1
CBD stones	ERC	Bleeding at papilla	1
Portal biliopathy with EHPVO	Removal of biliary stent	Rupture of choledochal varix	1
Klatskin tumour	PTC	Fistula between LHA and LHD	1

RHA: right hepatic artery, CHD: common hepatic duct, GDA: gastroduodenal artery, HJ: hepaticojejunostomy

CBD: common bile duct, EHPVO: extrahepatic portal vein obstruction, LHA: left hepatic artery, LHD: left hepatic duct

causing haemosuccus pancreaticus was present in 1 patient (Table 2).

The remaining 4 patients had bleeding following blunt abdominal trauma and injury to major vessels (Table 3).

In patients with iatrogenic injury the average interval between the injury and onset of bleeding was 29.5 (1–98) days. The average interval between intervention and bleeding was longer after surgical procedures (40 days, range of 10–98) than that after ERC or PTC (1.67 days, range 1–3). The cause of bleeding after surgery was pseudoaneurysm while there was no pseudoaneurysm after ERC or PTC. The average interval between LC and bleeding was 47(18–98) days (Table 4).

The commonest clinical presentation was gastrointestinal bleeding in 19 (95 percent) patients. Quinke's triad was present in only 3 (15 percent) patients (Fig. 1).

Management

The patients were managed according to our departmental algorithm (Fig. 2).

Nine patients were haemodynamically unstable at the time of presentation. After resuscitation and clinical diagnosis (including ultrasonography and upper gastrointestinal endoscopy), five patients underwent surgery, three underwent angiography and one underwent EST. Five patients had a clinical diagnosis of bleeding with associated conditions like liver necrosis with abscess, tear of the RHA and CHD, duodenal erosion, tear of LHA and LHD with hilar cholangiocarcinoma and misdiagnosis of gastric leiomyoma. Due to associated conditions these patients were directly taken up for surgery to stop the bleeding and treat the associated complications. The patient with post EPT bleeding had failure of EST and finally underwent surgery.

Eleven patients who were haemodynamically stable underwent CT angiograms of the upper abdomen. In all of them CT scan correctly diagnosed the cause of haemobilia. Six of these patients had associated conditions like duodenal erosion, bile leak, pseudocyst of the pancreas, extrahepatic portal venous obstruction with portal biliopathy, haemorrhagic cholecystitis and haemoperitoneum that warranted surgery. The remaining five patients had no

Table 2 Aetiology of spontaneous massive haemobilia

Spontaneous haemobilia (n=5)		
Underlying disease	Cause of bleeding	n
Stone in CHD	Pseudoaneurysm LHA	1
	Pseudoaneurysm RHA	1
Visceral aneurysm	GDA	1
	Multiple intrahepatic aneurysms	1
Pseudocyst with portal cavernoma	Pseudoaneurysm GDA eroding into pseudocyst	1

Table 3 Aetiology of accidental traumatic massive haemobilia

Accidental abdominal trauma (n=4, 20 percent)	
Site of bleeding	n
Laceration in segment V with tear in portal vein	1
Haematoma in R liver lobe with bleeding from RHV	1
Pseudoaneurysm in segment V branch of RHA	1
Pseudoaneurysm in segment VI branch of RHA with parenchymal laceration	1

Table 4 The time interval between intervention and bleeding

Procedures		n	Interval of bleeding (days)
Surgery	Laparoscopic cholecystectomy	6	18, 21, 28, 42, 75, 98
	Whipple's procedure	1	28
	Hepatic U-tube placement	1	10
Endoscopy/percutaneous	ERC and stone extraction	1	3
	Stent removal	1	1
	PTC	1	1

associated pathology and hence were subjected to conventional angiography and attempted embolisation.

Conventional angiography was attempted in eight patients. The site of bleeding could not be identified in one despite complete enhancement of the vascular tree and in another due to failure of cannulation of a tortuous common hepatic artery. In six patients the site of bleeding was correctly identified and TAE was attempted. In only two patients was the TAE successful in stopping bleeding. In three patients the bleeding persisted after placement of coils and they underwent surgery. In one patient selective cannulation of the anterior branch of the RHA could not be accomplished for placement of a coil.

Fig. 1 Clinical presentation of massive haemobilia

Surgery was performed on 18 patients. Failure of angiography and embolisation to control bleeding was the commonest indication for surgery accounting for 6 patients. Other indications were failure of EST and associated conditions that also required surgical management (Table 5).

The source of bleeding was arterial in 17 (85 percent) and venous in three (15 percent) patients (Table 6). Fourteen patients had bleeding from the aneurysm (either pseudo- or true aneurysm), three patients had bleeding from torn arteries and three had bleeding from torn veins. The commonest source of bleeding was a pseudoaneurysm in 12 (60 percent) patients, out of which an extra-hepatic pseudoaneurysm of RHA following LC was commonest. The venous bleeding occurred after blunt abdominal injury or with portal hypertension.

The procedures performed were aimed at controlling bleeding and providing a definite treatment of the underlying and associated conditions. Ligation or repair of the injured vessel was the commonest procedure performed (Table 7). Other procedures to control bleeding were right hepatectomy and lienorenal shunt. Nine patients also required a procedure to drain the biliary tree with either a T-tube or a hepaticojejunostomy. Concomitant procedures included drainage of a collection, duodenal repair, cystojejunostomy, distal gastrectomy, cholecystectomy and feeding jejunostomy.

Complications and mortality

Among the patients who underwent angiography, embolisation or EST none had procedure-related complications. Surgical complications occurred in seven patients (39%).

Fig. 2 Algorithm for management of massive haemobilia

Table 5 Indications for surgery in massive haemobilia

	Indications for surgery	n
Failure of angiography and/or embolisation		6
Failure of EST		1
Associated conditions	Pseudoaneurysm with duodenal erosion and abscess	2
	Tear of RHA with CHD	1
	Tear of LHA with cholangiocarcinoma	1
	Pseudoaneurysm and bile leak with abscess	1
	Pseudoaneurysm and chronic pancreatitis, pseudocyst	1
	Pseudoaneurysm with ductal stone and liver necrosis	1
	Rupture of choledochal varix with EHPVO	1
	Gangrenous cholecystitis	1
	Haemoperitoneum following blunt injury abdomen	1
	Misdiagnosed gastric leiomyoma	1

Table 6 Site of bleeding in patients with massive haemobilia

	Site of bleeding	n
Arterial (85 percent)	Extra-hepatic pseudoaneurysm RHA	5
	Intra-hepatic pseudoaneurysm RHA	4
	Extra-hepatic pseudoaneurysm LHA	1
	Pseudoaneurysm GDA	2
	Visceral true aneurysm of GDA	1
	Multiple intra-hepatic visceral aneurysms	1
	Tear RHA	1
	Tear LHA	1
	Bleeding from papilla	1
	Laceration segment 5 with bleeding from portal vein	1
Venous (15 percent)	Laceration segment 7 with bleeding RHV	1
	Ruptured choledochal varices	1

Table 7 Surgical procedures for control of bleeding

	Surgical procedure	n
Ligation of the artery with aneurysm	RHA	3
	LHA	2
	GDA	2
	Anteromedial branch of RHA	1
Repair of RHA		4
Right hepatectomy		2
Right hepatectomy with repair of portal vein		1
Proximal leiorenal shunt		1
Underrunning of papillary bleeder		1
Cholecystectomy		1

These included a bile leak, duodenal leak, duodenal stump blow out, intestinal leak with coagulopathy, refractory hypotension with renal failure, aspiration pneumonitis lead-

ing to ARDS and gastric fistula with sepsis. There was no rebleeding. All complications except gastric fistula were managed conservatively. The patient with the gastric fis-

tula was operated twice, once for repair of fistula and later for drainage of intra-abdominal abscesses. Four (22%) of the patients died. Three deaths occurred in those who had bleeding from torn arteries in their extra-hepatic course and one death in a patient who had a tear in the intrahepatic course of the right hepatic vein. None of the patients with either a pseudo- or true aneurysm died.

The average duration of hospital stay was 14.5 (range 1–50) days.

Statistical analysis

In patients who underwent surgery, the association between the type of bleeding and operative mortality was statistically assessed. There were four deaths out of six patients who had bleeding from tears in vessels and no deaths in 12 patients with a bleeding aneurysm. Due to the small sample size, Fisher's exact test was applied. The two sided 'p' value was 0.0049 that suggested a statistically significant association between bleeding from a non-aneurysmal cause and operative mortality.

Discussion

We saw a wide variety of causes of haemobilia most of which have been reported earlier as isolated case reports mainly from western countries. Iatrogenic trauma and particularly laparoscopic cholecystectomy is currently the commonest cause followed by spontaneous bleeding and accidental abdominal trauma. Goodnight and Blaisdell [12] in 1981 reported trauma in 50 percent cases of haemobilia with the ratio of accidental to iatrogenic being 2: 1. More recent reports however have shown a reversal of this etiological pattern. Hsu et al [13] in 2002 reported iatrogenic causes accounted for 12 out of 15 (80 percent) patients. Their series had no case of abdominal trauma. Green et al [1] in 2001 reported 66 percent to be iatrogenic, 29 percent spontaneous and 5 percent non-iatrogenic traumatic haemobilia. This pattern was also seen in our series. Others [1, 2, 14–18] have also reported the emergence of laparoscopic cholecystectomy as a cause of pseudoaneurysm and haemobilia. The previously reported concept of the Peter Pan syndrome in the context of LC [15] has gained further support based on observations of the present series. The other surgical procedures that caused pseudoaneurysms are Whipple's operation and hepaticojejunostomy with bilateral U-tube placement. Pancreatic head resections have a 12 percent reported incidence of bleeding from pseudoaneurysm [8] and hepaticojejunostomy is also a reported cause of hemobilia [19]. The non-surgical procedures like ERC and PTC though rare can cause massive bleeding. The percutaneous transhepatic needle procedures cause bleeding if they result in a fistula between an artery and the bile duct [4].

Spontaneous hemobilia may be due to visceral aneurysms or associated with underlying diseases like cholelithiasis and chronic pancreatitis with a pseudocyst. Pseudoaneurysms of the RHA, LHA or GDA is the cause of bleeding in these patients. As the hepatic ducts are in close proximity to the hepatic arteries, either surgical exploration or stones in the biliary tree may erode and cause haemorrhage [20].

Accidental blunt abdominal trauma is now a relatively rarer cause of massive hemobilia. The reported incidence of haemobilia after major blunt abdominal trauma in 1991 was around 3 percent or less [21, 22]. Surgery for hepatic lacerations can cause hemobilia by tearing of the bile ducts and vessels if the stitches are placed too deep or by the formation of cavities if stitches are placed too superficially [23].

The clinical presentation of haemobilia is nonspecific. Its commonest and most consistent feature is gastrointestinal bleeding. The preceding history of hepatobiliary surgery or accidental trauma should raise a high index of suspicion. The time interval between the iatrogenic trauma and bleeding depends on the cause of bleeding. It may take from 10 days to a few months for a pseudoaneurysm to form and bleeding may be instantaneous if vessels are torn by direct vascular trauma.

Algorithms for the management of massive hemobilia are in a phase of evolution. With the recent reports of successful nonoperative management the place of angiography with embolisation and surgery are not clearly defined. Despite advancement of techniques of nonoperative management, a subgroup of patients with massive bleeding may require operative treatment.

The CT scan should be the investigation of choice in stable patients who have a clinical diagnosis of haemobilia. In the present series it correctly identified the site of bleeding and other associated conditions whenever it was performed. To our knowledge this is the first series reporting the usefulness of CT scan in management of patients with massive hemobilia. The CT scan should follow the preliminary upper gastrointestinal endoscopy particularly if a liver parenchymal vascular cause is suspected to be the source of bleeding [12, 24]. This is helpful in deciding the further course of management. A stable patient having bleeding and associated conditions that require surgery for definitive management should undergo operation as a one time procedure for cure. In India the facilities and expertise of angiography and embolization are not widely available, hence the results of these procedures are also not as good as those reported in the western literature. We therefore recommend that only the patients who require control of bleeding and have no associated surgical conditions should be taken up for angiography and embolization.

Angiography is expected to detect a vascular abnormality in approximately 90 percent of cases with significant haemobilia [21]. The success rate of TAE as reported in most series is 80–100 percent [2–7, 9]. Most of them

however have evaluated both massive and minor haemobilia together. The reasons of failure of TAE in the present series could have been due to several factors. They include larger pseudoaneurysms with massive bleeding and multiple feeding vessels, intermittent bleeding, difficult anatomy and lack of expertise. In the present series angiography failed in two out of eight patients (25 percent) to localise the bleeding site and TAE failed in four out of six patients (67 percent) to stop the bleeding. The overall failures of transarterial embolization for haemobilia in most reports have ranged from 0 to 37 percent [1, 8, 9]. Although angiography is safer than surgery [25], it is not free of failures or complications. The complications after TAE include migration of coils and hepatic necrosis that has proved fatal in a few cases [8, 26].

In the current scenario where different reports show good results not only with TAE but even with blood transfusions alone [13], the role of surgery for massive hemobilia becomes unclear. We suggest that surgery should be offered to patients with associated surgical conditions and failed nonoperative treatment. We operated upon 18 patients, 11 (61 percent) for associated conditions (e.g. abscess, duodenal or biliary leak, portal hypertension, pancreatic pseudocyst, cholecystitis) and seven (39 percent) after the failure of nonoperative treatment. In the backdrop of our experience we propose that massive hemobilia should be managed as per the algorithm outlined above. Even if a patient is subjected to nonoperative intervention therapy, the surgical facilities must always be kept available for salvage in case of failure of nonoperative treatment.

Haemobilia is usually arterial in origin. Bleeding from venous fistula though rare [27], can occur with portal hypertension and accidental trauma. In our case of EHPVO a lienorenal shunt stopped the haemobilia and simultaneously decompressed the oesophageal varices. The bleeding due to laceration of the portal or hepatic vein following accidental trauma is inaccessible to nonoperative interventions and these are probably absolute indications for surgery.

The surgical procedures were tailored according to the requirement of an individual patient. The commonest procedure to control bleeding was ligation of the artery with an aneurysm with endoaneurysmorrhaphy being performed for major arteries. Hepatic resection was performed in cases of laceration or incomplete haemostasis after ligation of the feeding vessels.

The operative mortality was 22 percent and overall mortality was 20 percent. In 1972 the overall mortality for hemobilia was 25 percent [28]. By 1987 the mortality rate had fallen to 12 percent [6]. In 2001, mortality had further fallen to 2.25 percent [1]. All these reviews have included patients with minor hemobilia and correct mortality figures for massive bleeding hence cannot be ascertained. Hsu et al [13] in 2002 reported 27 percent mortality for massive hemobilia, mostly managed conservatively.

Despite the development of both nonoperative and operative techniques the mortality of haemobilia has remained unchanged over the last four decades. The patients should be managed according to resources available in individual settings. TAE or surgery are not alternatives; rather they should complement each other in the management of an individual patient.

Conclusion

Massive hemobilia constituted 9 percent of our patients with upper gastrointestinal bleeding who required surgery. A pseudoaneurysm resulting after LC was the commonest aetiology. The CT scan was a useful investigation to plan management. The mortality was significantly higher in non-aneurysmal bleeding. TAE should be attempted to control bleeding if there are no other conditions that require surgery. In India surgery perhaps should be considered early for associated conditions, venous bleeding, non-aneurysmal bleeding and failed TAE. Our overall mortality for patients with massive hemobilia was 22 percent.

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