Hepatic artery aneurysm following cholecystectomy

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Summary
A rare case is reported of an hepatic artery aneurysm developing 5 months after cholecystectomy. This presented as massive haemobilia as a result of the aneurysm communicating with the cystic duct stump. The diagnosis was delayed owing to the complicating features of Waldenström's macroglobulinaemia and jejunal diverticulosis.

Introduction
Hepatic artery aneurysms are uncommon (Guida and Moore, 1966), and the diagnosis is seldom made pre-operatively (Lataste, Albond and Melet, 1971). They may complicate biliary surgery, and indeed the diagnosis may be delayed when the patient presents with bile duct stones which could explain his symptomatology. A case is reported in which an aneurysm developed subsequent to biliary surgery, resulting in fatal haemobilia.

Case history
A 73-year-old retired male dairy worker was admitted as an emergency with epigastric pain and jaundice. He had previously been well apart from mild essential hypertension and a duodenal ulcer 11 years previously. His symptoms settled on conservative management and an i.v. cholangiogram revealed a dilated common bile duct but no opacification of the gall-bladder. He was discharged and readmitted 8 weeks later for elective cholecystectomy.

At laparotomy the anatomy was clearly displayed, and the gall-bladder found to contain multiple stones with a dilated common bile duct. A Meckel's diverticulum and marked jejunal diverticulosis were noted but the duodenum was normal with no evidence of ulceration. During dissection some haemorrhage occurred from the region of the porta hepatitis but this was controlled with a vascular stitch. The common bile duct was opened and explored following cholangiography, but no stones were found. As the lower end of the duct had not been clearly visualized, a choledochoduodenostomy was performed and the gall-bladder removed. The patient made an uneventful recovery.

He was readmitted 5 months later with 2 minor episodes of haematemesis. He had been well with no history of indigestion, and examination and gastroscopy were negative with no bleeding point found. One month later he had 3 further episodes of haematemesis and melaena and needed transfusion. Repeat gastroscopy again failed to reveal the site of bleeding and the duodenum and choledochoduodenostomy were clearly seen and appeared normal. Three days later he suffered massive haemorrhage resulting in cardiac arrest, and was resuscitated and taken to theatre. No lesion was found in the stomach or duodenum, although old clot was found in the stomach and small bowel, with some clot also seen in the lower common bile duct, which was thought to have regurgitated from the bowel. A truncal vagotomy and pyloroplasty were performed and the segment of jejunal diverticulosis excised. However, he bled again in the immediate postoperative period, which suggested that the bleeding site had not been dealt with adequately.

Arteriography was considered but deferred until he bled again as the success rate of diagnosis with this procedure is higher during active bleeding (Donahue and Nyhus, 1977). However, haematological examination revealed that the patient suffered from a heavy IgM paraproteinaemia which may have accounted for his tendency to bleed. Marrow examination revealed a slight increase in lymphocytes and a diagnosis of Waldenström's macroglobulinaemia was made. Plasma exchange was therefore arranged but before this could be undertaken, the patient suffered a further massive haemorrhage.

Emergency arteriography was then performed and revealed an aneurysm of the hepatic artery (Fig. 1). Immediate laparotomy was performed and the aneurysm was found to communicate with the cystic duct remnant, thus producing massive haemobilia. The patient continued to bleed excessively and, despite all resuscitative and surgical measures, died on the operating table.
Discussion

This case illustrates a very rare cause of haematemesis and melaena, in that a hepatic artery aneurysm developed following cholecystectomy and communicated with the cystic duct remnant, thus resulting in massive haemobilia. The diagnosis of haemobilia is often unfortunately delayed, as in this case, in which there were several complicating features. The history of duodenal ulceration, the presence of jejunal diverticulosis, and the findings of Waldenström's macroglobulinaemia, all complicated the clinical picture and delayed the diagnosis. It must be stressed therefore that haemobilia must be considered in all cases of obscure gastrointestinal haemorrhage, especially if accompanied by biliary colic and jaundice, although these latter features were absent in this case possibly because of the presence of the choledochoduodenostomy. The most common vascular lesion causing haemobilia is aneurysm of the hepatic artery (Harlaftis and Akin, 1977). The bleeding into the biliary tree may be variable, from a slow steady trickle causing melaena and secondary anaemia, to a rapid haemorrhage causing haematemesis and circulatory collapse (Harlaftis and Akin, 1977) and both these variations of blood loss were seen in this patient.

The development of an aneurysm of the hepatic artery following cholecystectomy is an extremely rare complication (Guida and Moore, 1966; Harlaftis and Akin, 1977). It appears to have developed since surgery as there was no evidence of aneurysmal dilatation at that time, in spite of good anatomical exposure. However, it is possible that it may have been related to the bleeding that was encountered during cholecystectomy, although it did appear to be in a different site, and the haemorrhage to have been well controlled by a vascular stitch. It may be argued that this manoeuvre produced a weakness in the vascular wall of the hepatic artery leading to aneurysmal dilatation, or even the formation of a false aneurysm. However, other previously reported cases developed this complication after an apparently straightforward cholecystectomy, and again the time interval was similar (Guida and Moore, 1966; Harlaftis and Akin, 1977). In their review of 80 cases in the literature, Guida and Moore (1966) found 23 were associated with biliary disease, and 2 of their own cases developed following cholecystectomy. It is thus impossible to draw any firm conclusions as to whether such aneurysms either predispose to, or are exacerbated by biliary disease, or indeed develop after such surgery. Nevertheless, this condition must be suspected in all cases of unexplained haemorrhage complicating biliary tract pathology (Hubens and De Schepper, 1979). The fact that the aneurysm communicated with the cystic duct in this present case explained the massive nature of the haemorrhage, which is extremely unlikely the haemorrhage usually seen with Waldenström's macroglobulinaemia.

Coeliac axis angiography is the only reliable method of diagnosing hepatic artery aneurysms (Nusbaum and Baum, 1963). These may require ligation and excision of the appropriate branch of
the hepatic artery (Taylor and Dawson, 1978) or graft reconstruction when affecting the main hepatic vessels (Goldman and Stone, 1974). Percutaneous embolization has also been utilized (Goldblatt et al., 1977), but where intra-hepatic rupture occurs, then ligation of the appropriate branch of the hepatic artery is necessary with resection of part of the liver. Accurate localization of the aneurysm is essential in order to plan the appropriate surgical procedure (Hubens and De Schepper, 1979). Simple proximal ligation is not always adequate (Erskine, 1973), and distal control is often difficult when the aneurysm is sited high in the porta hepatis or even within the liver. It is in this situation that resection of the involved liver segment may be necessary.

This case therefore illustrates many possible pitfalls in the diagnosis of haemobilia and is also presented as a rare complication following cholecystectomy. It demonstrates that delay in diagnosis can be fatal, as is seen in other cases (Guida and Moore, 1966; Hubens and De Schepper, 1979) and re-emphasizes the need for a high index of suspicion on obscure gastrointestinal haemorrhage, and a plea for earlier rather than later selective angiography.

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References