Upper gastrointestinal haemorrhage is a major medical emergency and accounts for approximately 7,000 admissions to hospitals in Scotland each year. Over the last 10 years there has been a number of improvements in diagnosis and conservative management of the condition, which significantly reduced the ratio of life-threatening cases requiring an emergency surgery. Despite these achievements surgical intervention or, if accessible, endovascular procedures must be undertaken as emergency actions, should conservative management fail. Vascular malformations of the duodenum are less frequent causes of upper GI bleeding. Duodenal varices found endoscopically occur in 0.4% of patients with portal hypertension (PHT) and are believed to be caused mainly by liver cirrhosis, idiopathic PHT, extrahepatic PHT, or previous surgical trauma. The duodenal bulb is their most common site, followed by the second portion of the duodenum. Forty per cent of patients with PHT have duodenal varices at angiography; however, their penetration unusually affects submucosa, hence no symptoms develop. Isolated bleeding duodenal varices are scarcely reported in literature, although present a significant surgical problem: massive haemorrhage combined with failure to identify them as a source has led to catastrophic outcomes with mortality rate of 40%. The case hereby presented is unique in several aspects. Duodenal varices were explored on emergency laparotomy rather than on prior endoscopies, which, performed by the same well-established endoscopists, were twice negative. This corresponds to the study by Cottam et al. stating that duodenal varices may not penetrate the submucosa, hence haemorrhages of their origin may even be more difficult to diagnose on endoscopy. Secondly, the haemorrhage here reported was undoubtedly a life-threatening condition that required a multidisciplinary team to be managed successfully. Along with Shirashi et al. we confirm that surgical ligation followed by the excision of duodenal / small intestinal varices may be an effective method of their management – both cases have been free of recurrence at 15 months postoperatively. In contrast to the study by Hashizume et al. the duodenal varices here presented were not associated with portal hypertension (PHT). Finally, duodenal varices located in the posterolateral aspect of the descending duodenum are less common as the majority of cases reported so far were of duodenal bulb location.

**Key words:** upper gastrointestinal haemorrhage, duodenal varices, oesophagogastroduodenoscopy, gastroduodenal resection
emergency surgery. A recent meta-analysis of 16 RCTs reported that combined endoscopic intervention (clipping / injection / thermal) for acute upper GI bleeding is associated with the need for surgery in only 7.6% of patients and with 2.6% mortality ratio (2). Despite these achievements surgical intervention or, if accessible, endovascular procedures must be undertaken as emergency actions should conservative management fail. According to the study by Rockall et al. among major causes of upper GI bleeding peptic ulcer is universally reported to be the most frequent accounting for 44% of cases found at emergency endoscopy. Further causes include oesophagitis (28%), gastritis/erosions (26%), erosive duodenitis (15%), varices (15%), portal hypertensive gastropathy (7%), malignancy (5%), Mallory-Weiss tear (5%) and vascular malformations (3%). Of note, in 20% of patients who underwent the study no endoscopic lesions were found (3).

Vascular malformations of the duodenum are less frequent causes of upper GI bleeding. This was reported by O’Mahony et al. on the basis of push enteroscopy (Olympus SIF-10 and SIF-10L) proceeded for upper GI bleeding in the seventeen consecutive patients who prior to push enteroscopy underwent routine OGD examination on which no abnormal lesions were identified. As push enteroscopy allows endoscopist to visualize up to 100 cm of the proximal intestine (the median length of small intestine intubated in the study was 60 cm), the following findings were reported: abnormalities were found in 10/17 patients (59%), six patients had small-intestinal vascular lesions: five had arteriovenous malformations (AVMs) of which one duodenal, and one patient had hereditary haemorrhagic telangiectasia. Two patients had lesions in stomach/duodenal bulb: one patient had a gastric AVM and one patient had duodenitis (4).

Duodenal varices found endoscopically occur in 0.4% of patients with portal hypertension (PHT) (5) and are believed to be caused mainly due to liver cirrhosis, idiopathic PHT, extrahepatic PHT, or previous surgical trauma (6). The duodenal bulb is their most common site, followed by the second portion of the duodenum (7). Forty per cent of patients with PHT have duodenal varices at angiography; however their penetration unusually affects submucosa hence no symptoms develop. Isolated bleeding duodenal varices are scarcely reported in literature, although present a significant surgical problem: massive haemorrhage combined with failure to identify them as a source has led to catastrophic outcomes with mortality rate of 40% (8, 9).

CASE REPORT

On the 27th of March 2008 a 53-year-old lady presented to A&E complaining on sudden overnight episode of coffee-ground vomiting and one day history of tarry stools. No other symptoms were voiced on admission. Her medical background included cirrhosis since 2003 with endoscopically confirmed oesophageal varices and duodenal ulcer. According to history taken and liver biopsy/HP examination the above were of alcoholic liver disease (ALD) origin. She had also been a heavy smoker of 18 years duration consuming twenty cigarettes a day. The only medication was one on oral asparin trice daily. She underwent open cholecystectomy in 1983 and two uneventful vaginal deliveries earlier. Family history was irrelevant. The patient was primarily assessed at 07:30, 27/02/2008 by the A&E doctor whose examination revealed patent airways, respiratory rate and chest auscultation were normal, regular tachycardia of 150/min, BP of 120/80 and GCS of 15/15 were noted.

On abdominal palpation the entire epigastrium was grossly distended, however bowel sounds were preserved and no peritoneal signs evident. Right sides epigastric scar was noted. Urgent blood tests were requested; their results at 07:49, 27/03/2008 were as follows; FCC 8 410/µL, HB 10.7 g/dL, PLT 312 000/µL, urea 57 mg/dl, creatinine 0.6 mg/dl, total bilirubin 1.5 mg/dl, AST 70 U/L, ALT 29 U/L, CRP 4.84 mg/l. After spending 35 minutes in A&E patient was transferred to the Department of Gastroenterology in view of further care. Fluid resuscitation, PPI, vitamin K, cycloamine and somatostatin had been administered intravenously. Urethral catheterization was performed. Urgent oesophagogastroduodenoscopy (OGD) (27/03/2008) showed no oesophageal varices, haemolyzed blood within the gastric fundus and body, pyloric longitudinal scar (presumed old ulcer site) with no signs of bleeding on its surface, irregular duodenal bulb and normal descending duodenum.
Results of follow up blood tests at 08:42, 27/03/2008 were: WCC 8 680/µL, HB 10.3 g/dL, PLT 301 000/µL, INR 1.27, K+ 4.4 mmol/l, Na+ 143 mmol/l, lipase 38 U/L, amylase 46 U/L, total protein 8.2 g/dL, GGTP 434 U/L. Throughout the first 6 hours of hospitalisation patient’s vital signs including urine output were acceptable, however coffee ground vomiting was gradually exacerbating and eventually some fresh blood within the vomiting appeared. The surgical review at that time advocated further conservative management. Second follow up FBC at 11:32, 27/03/2008 was: WCC 11 700/µL, HB 10.2 g/dL, PLT 185 000/µL.

Since haemathemesis and tarry stools persisted three units of blood and two units of fresh frozen plasma (FFP) were transfused between 14:55 and 23:35. Third follow up FBC at 22:38, 27/03/2008 was: WCC 10 500/µL, HB 7.5 g/dL, PLT 182 000/µL, whereas follow up FBC at 08:00, 28/03/2008 was: WCC 13 130/µL, HB 7.8 g/dL, PLT 192 000/µL. Follow up OGD, undertaken in the morning of the 28th of March 2008 by the same endoscopist who performed the previous endoscopy, showed again normal oesophagus, the cardia with an ulcer formed the previous endoscopy, showed again normal descending duodenum, and the trace of fresh blood spotted within the bulb and the descending duodenum. Despite best medical treatment being administered, during the day time of 28th March 2008 patient’s clinical status was deteriorating as more fresh blood vomiting occurred. Blood transfusion of three units of blood was undertaken between 11:30 and 16:00. Follow up FBC at 12:24, 28/03/2008 was: WCC 14 460/µL, HB 8.2 g/dL, PLT 183 000/µL, whereas follow up FBC at 14:27, 28/03/2008 was: WCC 11 840/µL, HB 9.5 g/dL, PLT 116 000/µL.

Patient’s overnight vital signs were stable including urine output of 1800 ml/24 hours, however in the morning of 29th March 2008 patient appeared drowsy and confused with blood pressure of 100/60 mm Hg and heart rate of 90/min. Further three units of blood were requested at 11:30, 29/03/2008 along with the second surgical review. At 13:45 a decision to proceed to emergency laparotomy was made. Blood pressure further dropped to 85/55 mm Hg while heart rate increased to 100/min. Blood test requested at 13:43, 29/03/2008 were: WCC 12 390/µL, HB 8.1 g/dL, PLT 122 000/µL, K+ 5.2 mmol/l, Na+ 138 mmol/L, INR 1.35, thus further two units of FFP were transfused prior to surgery. At 15:10 patient was transferred to theatre and generally anaesthetized.

Laparotomy via left medial incision revealed significant distension of stomach, duodenum and jejunum with dark-grey discolouration to their walls. Irregular multinodular surface of liver with no signs of portal hypertension was noted. No other intraabdominal pathology was evident. On gastrotomy a collection of blood clots within the stomach and some fresh bleeding inflowing from the duodenal site were observed. Despite thorough inspection of the gastric mucosa no source of bleeding was identified. Extension of gastrotomy towards duodenum was performed revealing a longitudinal scar of the mucosa on the posterior wall of the duodenal bulb likely representing an old ulcer. As oozing persisted Kocher Manoeuvre was initiated for further exploration. This was a breakthrough of the operation as major duodenal varices were found on the posterolateral aspect of the descending duodenum. The varices were adherent to the serosa, occupying the space in front of the inferior vena cava and the parietal peritoneum laterally. Diameters of the varices were ranging approximately from 1 to 10 millimetres, while their intramural penetration was difficult to establish (fig. 1, 2). Since the varices were the only finding that might potentially have caused the bleeding a decision to ligate them was made. Thorough ligation with subsequent excision of majority of the varices resulted in suppression of bleeding from the lumen of duodenum.

The Billroth I gastroduodenal resection followed by end-to-end hand-sutured anastomosis were performed with no intraoperative complications. During the operation which lasted for 4 hours and 50 minutes patient’s vital signs were stable; however additional four units of blood and two units of fresh frozen plasma were transfused along with infusion of 500 ml of crystalloids and 500 ml of colloids. After the operation patient was transferred to the Intensive Care Unit at the Department of General Surgery for further care. No respiratory support and catecholamines were required as patient’s initial post-op observations were
within acceptable ranges. Follow-up blood tests at 21:56, 29/03/2008 were: WCC 12 990/µL, HB 7.6 g/dL, PLT 75 000/µL, K+ 3.5 mmol/l, Na+ 142 mmol/L, serum albumin 1.8 g/dl, total protein 3.4, INR 1.47. Consequently both two units of albumins, two units of fresh frozen plasma and three units of blood were transfused by 08:00, 30/03/2008.

On the first post-op day patient’s haemodynamic status remained satisfactory. The gastric tube output was 400 ml of blood stained content, while two abdominal drains passed 1000 ml of blood-stained serous discharge. Follow-up blood tests at 10:13, 30/03/2008 were: WCC 9 400/µL, HB 8.1 g/dL, PLT 66 000/µL, K+ 3.3 mmol/l, Na+ 144 mmol/L, serum albumin 2.6 g/dl, total protein 3.9, INR 1.37, thus transfusion of three units of blood and two units of fresh frozen plasma along with further intravenous albumine supplementation was undertaken. FBC at 22:21, 30/03/2008 showed Hb of 11.6 g/dL, the other results were similar to the previous ones.

The second postoperative day (31/03/2008) was uncomplicated until evening when sudden respiratory failure occurred manifested by breathlessness which was confirmed on the arterial blood gas at 22:59, 31/03/2008 showing pO₂ 63.2 mm Hg, pCO₂ 30.2 mm Hg, O₂ sat 92.8%, pH 7.43, BE 1.9 mmol/l. Chest X-ray (31/03/2008) revealed vague bilateral shadowing over lower pulmonary areas with less extensive bilateral pleural effusions. This was managed by left sided pleural tap (100 ml of serous fluid was drained) and continuous oxygen supply of 15 L/min via external mask. A prophylactic antibiotic for presumed chest infection was given. Further care was uncomplicated and the asymptomatic patient was discharged from hospital on the 18th postoperative day. The follow-up angiogram dated (XX/XX/2008) showed no pathology of celiac axis and superior mesenteric artery in both arterial and venous phases of the imaging. Currently the patient remains asymptomatic and attends the regular out-patient appointments by the gastrology clinic.

**DISCUSSION**

The case hereby presented is unique in several aspects. Duodenal varices were explored on emergency laparotomy rather than on prior endoscopies, which performed by the same well-established endoscopists, were twice negative. This corresponds to the study by Cottam et al. (8) stating that duodenal varices may not penetrate the submucosa, hence haemorrhages of their origin may even be more difficult to diagnose on endoscopy. Secondly the haemorrhage here reported was undoubtedly a life-threatening condition that required a multidisciplinary team to be managed successfully. Not only the aggressive fluid resuscitation combined with transfusion of sixteen units of blood through four initial days of care was essential, but also the emer-
gancy surgery had to be undertaken to cease it. Along with Shirashi et al. (9) we confirm surgical ligation followed by excision of duodenal / small intestinal varices may be an effective method of their management – both cases have been free of recurrence at 15 months postoperatively.

In contrast to the study by Hashizume et al. (5), the duodenal varices here presented were not associated with portal hypertension (PTH). Although the patient suffered from long standing liver cirrhosis, PTH was ruled out on the basis of both thorough intraoperative exploration and follow-up angiogram. Finally, duodenal varices located in the posterolateral aspect of the descending duodenum are less common as majority of cases so far reported were of duodenal bulb location (7).

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