Impending Cholecystoduodenal Fistula As A Cause For An Unusually-Sited Bleeding Duodenal Ulcer

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Citation

Abstract
Background: Peptic duodenal ulceration (DU) is a common cause for upper gastrointestinal bleeding, and usually affects the duodenal bulb. Post-bulbar DU is unusual and may rarely be caused by a cholecystoduodenal fistula (CCDF).

Methods & Observations: We report a 66-year-old patient who presented with haematemesis and was found to have a post-bulbar bleeding DU that was managed by endoscopic intervention. Seven weeks later, he presented with a gallstone ileus, and was managed successfully with laparoscopy enterolithotomy. A barium study demonstrated a CCDF at the site of the previously detected DU.

Conclusion: Impending CCDF secondary to a large gallstone ought to be considered as a cause for an unusually sited bleeding DU in elderly patients. Consideration may then be given to an elective management of the cholecystolithiasis with percutaneous lithotripsy and dissolution therapy in the surgically unfit patient or with a laparoscopic cholecystectomy and closure of the CCDF.

INTRODUCTION
The most common causes of upper gastrointestinal (GI) bleeding include peptic ulceration, oesophageal varices, reflux oesophagitis, Mallory-Weiss tear, gastritis and malignancy. Duodenal ulceration (DU) is a manifestation of peptic ulcer disease and, by enlarge, is a disease of the duodenal bulb (Al-Bahrani 1980). Rarely, however, a peptic DU may occur more distally in a post-bulbar location, and may be attributed in some of the patients to Zollinger-Ellison syndrome (gastrinoma). Other rare causes for post-bulbar DU may include Crohn's disease, tuberculosis, and malignancy.

The development of a cholecystoduodenal fistula (CCDF) in patients with large gallstones is a rarely recognised cause of upper GI bleeding. We report an elderly patient who presented with a bleeding DU that was unusually sited at the junction of the first and second parts of the duodenum and was later shown to be secondary to a recently formed CCDF.

CASE REPORT
A 66-year-old disabled man with a right-sided hemiplegia and dysphonia secondary to a cerebrovascular accident 19 years previously, presented to the Accident and Emergency (A&E) Department with haematemesis and collapse. His family gave a past history of repeated vomiting after meals over the preceding six months, for which he was seen once in the A&E Department and treated conservatively for a presumed Mallory-Weiss tear. He has had an appendicectomy in early adulthood. His pulse and blood pressure on admission were 90/min and 105/70 mmHg. He had haemoglobin of 7.8 g/dl, a white cell count of 14.6 x10^9/l, a serum amylase of 255 u/l (normal <100 u/l), and a normal clotting screen.

He underwent an urgent endoscopy after appropriate resuscitation with intravenous fluids and blood transfusion and was found to have a 1.5 cm diameter posterior duodenal ulcer at the junction of the first and second parts of duodenum with a visible bleeding vessel at its edge. The ulcer was injected with adrenaline 1:10000 and the bleeding stopped. An antral biopsy for Campylobacter-like organisms (CLO test) was negative. He was placed on proton pump inhibitor therapy. He re-bled the following day, became hypotensive and dropped his haemoglobin to 5.8 g/dl.

Following resuscitation, a re-endoscopy was performed under general anaesthesia with view to progress to a laparotomy if necessary. The bleeding duodenal ulcer was however controlled with a re-injection of adrenaline. He was treated thereafter for a right-sided basal pneumonia, was
transferred to the Stroke Rehabilitation Unit 17 days after admission, and was discharged home 16 days later.

He was readmitted 16 days after discharge with a 3-day history of persistent vomiting, central abdominal pain and absolute constipation. A plain abdominal radiograph showed dilated small bowel loops with a suspicion of an air bubble in the liver. A laparoscopy was carried out and revealed a 4-cm gallstone lodged at mid-jejunum causing a proximal small bowel obstruction. The stone was removed laparoscopically through an enterotomy.

He was subsequently treated for a further episode of pneumonia, investigated for a swallowing disorder, and was transferred to the Neurology rehabilitation ward home on the 18th postoperative day. A post-surgery barium meal demonstrated a CCDF at the junction of the first and second parts of the duodenum (Figures 1A & 1B), and abdominal ultrasonography showed gas in the gallbladder and biliary tree but no further gallstones.

**Figure 1**
Figures 1A & 1B: Barium swallow post-laparoscopic enterolithotomy showing a cholecystoduodenal fistula (arrow) at the junction of the first and second parts of the duodenum; the site of the bleeding duodenal ulcer that was previously detected at endoscopy

**DISCUSSION**
Upper GI bleeding secondary to the formation of a CCDF has been reported previously in an elderly patient who was found to have a gallstone ulcerating into the second part of the duodenum, and in five others with large gallstones that obstructed the duodenum (Bouveret's syndrome). The stones were removed surgically through a laparotomy and enterolithotomy or duodenotomy in most patients, and via ultrasound-guided extracorporeal shockwave lithotripsy followed by endoscopic extraction in one patient. In another elderly patient, haematemesis resulted from the development of a cholecystogastric fistula that was followed 6 months later with a fatal gallstone ileus. In the elderly patient of our report, the impending formation of a CCDF was associated with significant upper GI bleeding from an ulcer unusually sited at the junction of the first and second parts of the duodenum that was followed some 7 weeks later with the development of gallstone ileus; the stone was retrieved at a laparoscopic enterolithotomy.

The possibility of an impending CCDF as a cause for an upper GI bleed may be entertained in an elderly patient with an endoscopic finding of a post-bulbar duodenal ulcer. Gallstones that may lead to the development of a CCDF are usually large and often lead to gallstone ileus; a condition commonly observed in the elderly and carries a considerable mortality risk (15-18%). The point of ulceration invariably occurs distal to the duodenal bulb and proximal to the third part of the duodenum, though ulceration into the duodenal...
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Impending CCDF is a rare and often unrecognised cause of upper GI bleeding in the elderly and may be suspected at endoscopy when a duodenal ulcer is detected distal to the duodenal bulb or within the second part of the duodenum. Confirmation of the diagnosis by ultrasonography and barium study may enable a timely treatment of the culprit, often large, gallstone with either lithotripsy and litholytic therapy or a laparoscopic cholecystectomy and closure of the fistula and avoid the complication of gallstone ileus.

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bulb, stomach, may occasionally occur and present with upper GI bleeding. The diagnosis may be supported or confirmed on abdominal radiography and ultrasonography, which may readily demonstrate a large calcified gallstone and possibly air within the biliary tree if the fistula has indeed formed. A barium study would reveal an established fistula.

It is worth investigating the possibility of an impending CCDF in such patients as a timely intervention might spare an urgent surgery under less favourable clinical conditions in these elderly patients. The gallstone may be amenable to treatment with extracorporeal shock wave lithotripsy and dissolution therapy in the infirm and unfit patient. Indeed, gallstone ileus is typically caused by large gallstones, which are more amenable to lithotripsy. In patients undergoing this mode of therapy the recurrence rate of gallstone formation is small during the short-term follow up. Endoscopic lithotripsy may be feasible. Alternatively, a timely laparoscopic cholecystectomy in the favourable-risk patient with closure of the CCDF would be feasible.

Impending CCDF is a rare and often unrecognised cause of upper GI bleeding in the elderly and may be suspected at endoscopy when a duodenal ulcer is detected distal to the duodenal bulb or within the second part of the duodenum.
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