Unusual presentation of more common disease/injury

Upper gastrointestinal bleeding: gallstone-induced auto-sphincterotomy

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Summary

A 67-year-old gentleman with no significant medical history of note presented with sudden onset of epigastric pain, coffee ground vomiting and passing black tarry stool. A series of investigations including blood tests, ultrasound scan, CT abdomen and pelvis with contrast and endoscopy failed to reveal any site of active bleeding. The mystery remained and the patient continued to have upper gastrointestinal bleeding. A second CT abdomen and pelvis with contrast was carried out and showed evidence of contrast extravasation into the duodenum (figure 3). An exploratory laparotomy showed no obvious site of haemorrhage and a loop jejunostomy was performed. The diagnosis of gallstone-induced auto-sphincterotomy was only made, using gastroscope via jejunostomy, when a big gallstone was found in the third part of the duodenum and the papilla was ruptured (figure 5).

BACKGROUND

Gastrointestinal (GI) bleeds can be life threatening and pose a diagnostic dilemma in the event that an oesophagogastroduodenoscopy (OGD) and colonoscopy do not reveal the cause of bleeding. Based on a literature search (Medline), there have been no case reports of a gallstone performing an auto-sphincterotomy or causing trauma at the ampulla of Vater, precipitating a massive upper gastrointestinal (UGI) bleed. In our patient a gallstone was observed in the third part of the duodenum at a relook endoscopy via a jejunostomy. No other cause for the bleeding was identified and there was no evidence of the gallstone having fistulated into the duodenum.

CASE PRESENTATION

On 14 December 2011, a 67-year-old gentleman with a background of angina pectoris, arthritis, colonic polyp (2009) and intermittent claudication (2001) was found collapsed in the toilet and he was taken to the Accident and Emergency Department. He was in hypovolaemic shock with a blood pressure of 80/40 mm Hg, pulse of 50, temperature of 34°C and was resuscitated appropriately. Following resuscitation, the patient was transferred to the acute admissions unit for definitive management. A collaborative history was taken from the patient's wife as he was unable to communicate sufficiently. The patient had been complaining of epigastric pain for 3–4 days, had two episodes of coffee ground vomiting and had been passing black tarry stool. He had no previous episodes of GI bleeding. On examination, the patient appeared to be pale and clammy. Glasgow Coma Scale was 10/15 on admission and 15/15 after resuscitation. The examination findings were as follows: epigastric tenderness with guarding; absence of rebound tenderness; negative Murphy's sign; negative shifting dullness; presence of bowel sound; fresh blood in nostrils and per rectum and bibasal crepitations. The patient continued to have multiple episodes of per

rectal (PR) bleeding and melaena during his admission while a series of investigations were being conducted.

INVESTIGATIONS

The blood tests revealed a sodium of 143 mmol/l, potassium of 5.8 mmol/l, urea of 8.3 mmol/l, creatinine of 101 mmol/l, normal liver function tests, C reactive protein 6 mg/l, haemoglobin of 11.7 g/dl and a white cell count of $11.7 \times 10^9/l$. Ultrasound scan did not reveal gallstones or dilatation of the common bile duct. OGD on 14 December 2011 showed mild erythematous/exudative gastritis and mild-to-moderate duodenitis (figure 1). The examination to the second part of the duodenum did not find any source or signs of recent upper GI haemorrhage. A CT scan of the abdomen and pelvis with contrast on 15 December 2011 revealed no active contrast extravasation in the small or large bowel. Multiple calcific mesenteric nodes were noted. No other significant findings

Figure 1 Oesophagogastroduodenoscopy (14/12/11): Mild erythematous/exudative gastritis and mild-to-moderate duodenitis. There was no evidence of an active bleed or ulcer.

Figure 2 Sigmoidoscopy (15/12/11): The evidence was of ongoing bleeding and melaena.

were noted on other abdominal organs. A Flexible Sigmoidoscopy on 15 December 2011 showed evidence of ongoing bleeding and melaena (figure 2). Therefore, colonoscopy, with successful caecal intubation was carried out the next day which revealed diverticulosis. No active bleeding site was identified. However, dark blood and melaena were seen in the lumen up to the caecum. A repeat OGD was considered but clinically the ongoing bleeding was felt to be 0.5–1 ml/min, due to transfusion requirements, and hence a second CT abdomen and pelvis with contrast was carried out on 18 December 2011. This scan successfully showed evidence of contrast extravasation into the second and third part of the duodenum and into the proximal aspect of jejunum which were suggestive of active bleeding in these areas (figure 3). No other obvious areas of active contrast extravasation noted in other parts of bowel. In view of the ongoing episodes of PR bleed and melaena, exploratory laparotomy was carried out on the same day to investigate the GI bleed. At the laparotomy, blood was seen in the small bowel up to the duodenum. A jejunotomy was performed and a gastroscope passed retrograde into the duodenum. An area of redness with a small pool of blood was noted distal to the papilla. This was aspirated and no active bleeding was seen. Careful examination of the gall bladder and the duodenum was ‘Kocherised’ to release it from the retroperitoneal attachment to look for any external signs of ulcers or tumours. No source for the haemorrhage was seen (figure 4).

The mystery remained. An old-fashioned surgical technique—a loop ‘jejunostomy’ was done and the abdomen closed. This was to allow a gastroscope to pass directly via the jejunum into stomach and it would differentiate between an upper and lower GI bleed. Nonetheless, no active bleeding was identified. There was no further bleeding seen from either limbs of the jejunostomy for the next 3 days.

An endoscopy via jejunostomy was conducted on 21 December 2011 to identify the cause of bleeding. The distal lumen endoscopy did not find any blood in the lumen and on proximal endoscopy via the jejunostomy a large gallstone was seen in third part of duodenum which was removed with a Roth net (figure 5). This was 9 mm in diameter. Since the gallstone was found in the duodenum, the papilla in the duodenum was carefully examined, and was found to be ruptured presumably by the large gallstone passing through it. No active bleeding was observed. Therefore, by eliminating all other causes, the bleeding was most likely due to the passage of the large gallstone through the papilla, leading to an auto-sphincterotomy. The jejunostomy was reversed a few days later (28 December 2011) once we were certain that there was no further bleeding.

DIFFERENTIAL DIAGNOSIS

Differential diagnosis of upper GI bleeding in this case includes peptic ulcer disease, gastritis, duodenitis, oesophagitis, GI malignancy, oesophageal or gastric

Figure 3 CT abdomen and pelvis with contrast (18/12/11): this scan showed evidence of contrast extravasation into the distal aspect of the duodenum and into the proximal aspect of jejunum which were suggestive of active bleeding in these areas. No other obvious areas of active contrast extravasation noted in other parts of bowel.

Figure 4 Gastroscopy via jejunostomy (21/12/11): Looking at pylorus from duodenum. No active bleeding was identified.

varices, Mallory-Weiss tear, bleeding disorder, angiodysplasia and aortoenteric fistula.

TREATMENT

The patient was resuscitated in the emergency department with 500 ml normal saline and 250 ml a colloid solution. 100% oxygen was administered to the patient. He was transferred to acute medical unit once stabilised, where a further 2 l of normal saline and two units of blood were administered. A series of investigations were carried out over the subsequent days in an attempt to locate the source of the bleeding. On 18 December 2011, he collapsed on the ward and a repeat contrast CT scan was carried out. An exploratory laparotomy was conducted which revealed large amount of blood in the small bowel. He received five units of blood intraoperatively due to a haemoglobin of 6.6 g/dl.

OUTCOME AND FOLLOW-UP

The patient made a full recovery and did not have any further episodes of bleeding since his closure of jejunostomy. He is being followed up in the outpatients clinic and a follow-up ultrasound scan was planned.

DISCUSSION

An UGI bleed may present in one of several ways; with haematemesis, coffee ground vomiting, melaena, hematochezia or fresh bleeding per rectum in a massive haemorrhage.

It is estimated that up to 10 000 admissions, incurring a total cost of at least £20 million, are attributed to UGI bleeds.¹

The incidence for non-variceal bleeds is 84.6 per 100 000 population and 2.83 per 100 000 population for variceal bleeds.¹

In the UK UGI bleeds are four times more common than lower GI (LGI) bleeds, with a male-to-female ratio approaching 1 : 1 however some studies still report a slight male prevalence.

Mortality rates are 10% overall.² UGI bleeding accounts for over 9000 deaths a year in the UK³ and over 1000 deaths per year could be prevented in England.¹

In order to classify a bleed as UGI the anatomic cut-off is considered to be the ligament of Treitz. Bleeding from a point distal to this is classified as a LGI bleed.

Common causes of UGI bleeds include: oesophagitis, gastritis, oesophageal/gastric varices, oesophageal/gastric/duodenal ulcers (including Cushing/Curling ulcers and Cameron lesions), Mallory-Weiss tears, oesophageal/gastric malignancies, benign gastric tumours, Boerhaave's syndrome, portal gastropathy and iatrogenic.

Other associations include: H Pylori, Non Steroidal Anti-inflammatory Drugs, Aspirin, other antiplatelets/anticoagulants and Selective Serotonin Reuptake Inhibitors.

Less common causes include: Dieulafoy's lesions, gastric antral vascular ectasia, Von Willebrand disease, Zollinger-Ellison syndrome, angiectasias, Osler-Weber-Rendu syndrome, haemobilia, aorto-enteric fistulae, pancreatic pseudoaneurysm, haemosuccus pancreaticus, Ehlers-Danlos syndrome, Superior mesenteric artery syndrome.

Other rare causes have been recorded in the literature and these include:

1. Periapillary⁴/Jejunal⁵ Dieulafoy's lesions
2. Life-threatening bleeding from duodenal varices due to pancreatic arterio-venous malformation.⁶
3. Bleeding from duodenal ulcer in a patient with bilio-pancreatic diversion.⁷
4. Pyloric giant Brunner's gland hamartoma as a cause of both duodenojejunal intussusception and obscure GI bleeding.⁸
5. Complicated duodeno-biliary fistula in bleeding duodenal ulcer: case report and literature review.⁹
6. Neurofibroma of the small intestine as a cause of massive bleeding from the GI tract.¹⁰

Figure 5 Gastroscopy (21/12/2011): A large gallstone was seen in third part of duodenum and this was removed using a Roth net. There was no bleeding at this time. The papilla was evaluated and clear bile was seen entering the duodenum.

7. Extensive gastric heterotopia of the small intestine resulting in massive GI bleeding, bowel perforation, and death.¹¹
8. Pseudoxanthoma elasticum. A rare cause of massive GI haemorrhage.¹²

Poor prognosticators of outcome and high risk for rebleeding should be considered in the following situations: age greater than 60 years, multiple comorbidities, variceal bleeding, coagulopathy, shock, bright red bleeding, large volume blood transfusions or repeated transfusions and the need for emergency surgery. Endoscopy findings which indicate a higher risk of rebleeding and an increased death rate include: active bleeding/oozing, bleeding from a large ulcer and a visible/spurting vessel.

The Glasgow-Blatchford, Forrest, Baylor and Rockall are classifications associated with UGI bleeds. These are used to help determine patients appropriate for endoscopy; to classify the lesion in an attempt to predict mortality risk, the risk of poor outcome and the possibility of rebleeding.

There are several options that may follow an initial period of resuscitation, the gold standard being an OGD. Therapeutic options are available at OGD and may include: banding of varices, injection of epinephrine/sclerotherapy, other haemostatic materials (biological glue), endoscopically applied clips, electrocautery, argon plasma coagulation, laser photocoagulation and tamponade using constant probe pressure.

Mesenteric angiography is considered when an OGD has failed to identify the source of the bleeding. It is most accurate with a bleeding rate of more than 0.5–1 ml/min.¹³ If the mesenteric angiogram has successfully localised the point of bleeding, then rarely selective embolisation is considered. The mesenteric angiogram and selective embolisation is not available at all institutions due to the need for highly specialised interventional radiologist and equipment. A repeat OGD is the alternate investigation at this point. This may also provide useful information in the event that surgery becomes necessary. Anti-fibrinolytics, recombinant Factor VII and other pharmacotherapy may be considered.

The difficulty occurs when the patient continues to bleed and the investigations have not revealed a cause. At this point, surgery is considered however this is no easy feat as the bleeding point needs to be located accurately in order to minimise the extent of the bowel resection.

Some of the indications for surgery are as follows: massive haemorrhage/haemodynamic instability despite resuscitative efforts, failure of medical therapy/endoscopic intervention with persistent recurrent bleeding, prolonged/continuous bleeding requiring recurrent/daily blood transfusions and recurrent bleeding leading to instability/deterioration.

Enterotomies have been well documented as an option and this can be followed by on table enteroscopies in order to locate the bleeding point.

Another method described is the use of atraumatic bowel clamps to section off a length of bowel and thus the entire length of the small bowel can be sequentially examined.

Complications of gallstones are well documented and some include pancreatitis, cholangitis, gallstone ileus, Mirizzi's syndrome, Bouveret's syndrome and GI bleeding; however none of these had been due to a non-therapeutic sphincterotomy.

While therapeutic sphincterotomies can cause bleeding there is no documentation of a gallstone resulting in a 'natural' sphincterotomy and hence causing a UGI bleed.

Learning points

- ▶ Although 'common things occur commonly', we do have to bear in mind the less common presentations, which is why it is important to report cases and read the literature. When encountering a rare presentation like ours remember the Sherlock Holmes quote: '... when you have eliminated the impossible, whatever remains, *however improbable*, must be the truth'.¹⁴
- ▶ Establishing a diagnosis (upper gastrointestinal (UGI) vs lower GI bleed) is not always straightforward and neither is locating the source of the bleeding within the GI tract, hence the clinical picture needs to be reassessed and investigations may need repeating.
- ▶ Lateral thinking is required, for example the formation of the jejunoscopy, and when there is no straightforward solution, it is paramount to do what is safest and plan to relook/reassess

Competing interests None.

Patient consent Obtained.

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