On-line Case Report

A case of obscure gastrointestinal bleeding in a teenager

N ABBASSI-GHADI, K BARTLAM, S RASHEED, T HOLME

Department of Surgery, Lister Hospital, Stevenage, UK

ABSTRACT

In this report, we discuss a case of obscure gastrointestinal bleeding (OGIB) in an 18-year-old man, where oesophagogastroduodenoscopy (OGD) and colonoscopy proved inconclusive in determining a source of bleeding. On day 14 of admission, a laparoscopy was performed, identifying a mass 2 feet from the ileocaecal junction. This was thought initially to be a Meckel's diverticulum and the patient underwent a laparotomy and a small bowel resection. Surprisingly, the histology revealed a benign gastrointestinal stromal tumour (GIST) of the ileum, rare in such a young person. Given the diagnostic delay, we propose a simple algorithm for the diagnostic management of a teenager presenting with a significant ongoing gastrointestinal bleed.

Keywords: Obscure gastrointestinal bleeding (OGIB) – Gastrointestinal stromal tumour (GIST) – Meckel's diverticulum

The term gastrointestinal stromal tumour (GIST) was originally used to describe all mesenchymal gastrointestinal tumours, as a collective term used to include tumours of neural and muscle origin.¹ GISTs have an incidence of 15 per million, more commonly occur in middle-aged and older people with approximately equal sex distribution and have an overall 5-year survival of about 35%.¹ The most commonly affected site is the stomach, followed by the small intestine, colon and rectum.² A recent study on the epidemiology of GISTs including 1458 cases, demonstrated that these tumours commonly occur in those over the age of 40 years. No cases were reported in those under the age of 20 years.³

GIST cells closely resemble the interstitial cells of Cajal (ICC), or gastrointestinal 'pacemaker cells'. ICCs arise from precursor mesenchymal cells and express the trans-membrane tyrosine kinase receptor, KIT. Gene mutations in these cells result in KIT protein overexpression, causing

neoplastic transformation and development of gastrointestinal stromal tumours. This may be detected by immunohistochemistry using the antibody CD117, which allows for accurate diagnosis of GISTs and differentiation from non-GIST tumours of the gastrointestinal tract.⁴

Surgical resection of the tumour is the mainstay of treatment for primary disease. Lymph node dissection is not recommended since metastatic spread is rare.

The tyrosine kinase inhibitor, Imatinib Mesylate (Glivec®) has also been used successfully in those cases not amenable to curative surgery or for recurrent and metastatic disease.⁴

Whilst there is an abundance of literature on the causes and diagnostic management of per rectal bleeding in adults, very little has been documented with regards to young people. We report a case of GIST arising in the ileum, 2 feet from the ileocaecal junction, causing significant gastrointestinal bleeding in an 18-year-old.

Correspondence to: T Holme, Consultant Surgeon, Lister Hospital, Stevenage SG1 4AB, UK

T: +44 (0)1438 314333; E: tom.holme@nhs.net

Case report

An 18-year-old man was admitted under the general surgical team with a 3-day history of dark-red per rectal bleed mixed with his stools, central colicky abdominal pain and three episodes of vomiting with no haematemesis. He had no significant previous medical history, was on no regular medication and he had no allergies.

Clinical examination was unremarkable except for a tachycardia of 120 bpm. Rigid sigmoidoscopy revealed stools mixed with dark-red blood tracking down beyond the rectosigmoid junction. Blood tests showed a haemoglobin level of 8 g/dl (MCV 84 fL) with normal inflammatory markers, U&Es, LFTs, clotting and amylase. AXR showed a normal gas pattern of the bowel.

The following day, an oesophagogastroduodenoscopy (OGD) up to the distal duodenum was performed, but was normal throughout. A colonoscopy on the fifth day of admission revealed blood throughout the colon and distal

Figure 1 Laparotomy exposing the GIST 2 feet from the ileocaecal junction.

ileum. A Meckel's 99m -technetium pertechnetate isotope scan on the ninth day of admission was unremarkable. Although he was stable, he continued to bleed intermittently and had

Figure 2 Diagnostic algorithm for on-going gastrointestinal bleeding in a teenager.

received a total of 12 units of blood. A decision was made to proceed with a diagnostic laparoscopy.

Laparoscopy on day 10 revealed a 30-mm mass 2 feet from the ileocaecal junction (Fig. 1), which was thought to be a Meckel's diverticulum. He had a lower mid-line laparotomy followed by a small bowel resection of the lesion with primary end-to-end anastomosis and an appendicectomy. He made an uneventful recovery with no further bleeding and was discharged from hospital 18 days after admission.

Surprisingly, histopathological examination revealed a benign GIST with clear resection margins. The tumour stained positive for CD117 and smooth muscle antibody (SMA). The appendix was normal. He was then referred for follow-up at a tertiary centre with a special interest in GISTs.

Discussion

GISTs are a rare cause of gastrointestinal tract bleeding. The most common presentation of GISTs is bleeding (50%), followed by abdominal pain (20–50%) and intestinal obstruction (20%).²

The most common causes of upper gastrointestinal bleeding in children older than 12 years are oesophageal varices and gastritis. Lower gastrointestinal bleeding is most commonly caused by polyps, inflammatory bowel disease, infectious diarrhoea and vascular anomalies.⁵ The majority of these disorders would usually be diagnosed by standard OGD and colonoscopy.

This report highlights a rare case of GIST causing on-going bleeding in a teenager, which resulted in diagnostic difficulties. In fact, it took a total of 10 days to diagnose the source of bleeding after he had received a total of 12 units of blood. The OGD and colonoscopy were performed within 5 days of admission but did not provide a diagnosis.

Bleeding is considered to be obscure if a source of blood loss is not apparent from evaluation of the upper gastrointestinal tract and colon. Therefore, for a case of OGIB, one must be able to image the small bowel effectively to establish a cause. Diagnostic limitations of imaging the small bowel create problems in accurate, early diagnosis. The judicious use of video capsule

endoscopy (VCE) and double balloon enteroscopy (DBE) could be useful diagnostic tools. A prospective study of patients with OGIB demonstrated a diagnostic detection rate of 80% and 60% for VCE and DBE, respectively,⁶ and these methods can be employed to detect GISTs presenting with OGIB.

Other diagnostic methods to consider include contrast-enhanced computed tomography (CT) and CT angiography although these introduce the associated risks of radiation exposure in the young patient. Standard angiography and CT angiography have diagnostic limitations and require on-going bleeding. A Meckel's scan has high sensitivity for a Meckel's diverticulum except when there is active bleeding, as it induces a washout of the radioisotope in the Meckel's diverticulum.

Diagnostic laparoscopy may be used as a definitive method in OGIB. In a report of 22 cases of OGIB, laparoscopy successfully detected the site of bleeding in 100% of cases.⁷

In view of the evidence above and our own experience, we have created a simple algorithm regarding diagnostic management in a teenager with on-going gastrointestinal bleeding (Fig. 2).

References

1. DeMatteo RP, Lewis JJ, Leung D, Mudan SS, Woodruff JM, Brennan MF. Two hundred gastrointestinal stromal tumors: recurrence patterns and prognostic factors for survival. *Ann Surg* 2000; **231**: 51–8.
2. Mucciarini C, Rossi G, Bertolini F, Valli R, Cirilli C, Rashid I *et al.* Incidence and clinicopathologic features of gastrointestinal stromal tumours. A population-based study. *BMC Cancer* 2007; **7**: 230.
3. Tran T, Davila JA, El-Serag HB. The epidemiology of malignant gastrointestinal stromal tumours: an analysis of 1,458 cases from 1992 to 2000. *Am J Gastroenterol* 2005; **100**: 162–8.
4. O'Leary T, Berman JJ. Gastrointestinal stromal tumors: answers and questions. *Hum Pathol* 2002; **33**: 456–8.
5. Fox VL. Gastrointestinal bleeding in infancy and childhood. *Gastroenterol Clin North Am* 2000; **29**: 37–66.
6. Hadithi M, Heine GD, Jacobs MA, Van Bodegraven AA, Mulder CJ. A prospective study comparing video capsule endoscopy with double-balloon enteroscopy in patients with obscure gastrointestinal bleeding. *Am J Gastroenterol* 2006; **101**: 52–7.
7. Ba MC, Qing SH, Huang XC, Wen Y, Li GX, Yu J. Application of laparoscopy in diagnosis and treatment of massive small intestinal bleeding: report of 22 cases. *World J Gastroenterol* 2006; **12**: 7051–4.