Jejunal GIST: Hunting down an unusual cause of gastrointestinal bleed using double balloon enteroscopy. A case report

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**A B S T R A C T**

**INTRODUCTION:** Gastrointestinal stromal tumours (GISTs) are the most common mesenchymal neoplasms of the alimentary tract but accounts for only 0.1–3% of all gastrointestinal neoplasms. The most common presentation of GISTs is acute or chronic gastrointestinal bleeding, in which the patient presents with symptomatic anaemia.

**PRESENTATION OF CASE:** With that in mind, we describe a 66-year-old man who presented with recurrent episodes of obscure gastrointestinal bleeding for two years. Video capsule endoscopy (VCE) showed several small telangiectasias in the proximal small bowel. Oral route double-balloon enteroscopy (DBE) revealed abnormal mucosa 165 cm from incisor with central ulceration and vascular component. He subsequently underwent surgical excision. The histopathological report confirmed the diagnosis of GIST arising from the jejunum. During his clinic follow up, he remains symptom-free with no evidence of recurrence.

**DISCUSSION:** The diagnosis of bleeding small intestine GISTs can be challenging as these are inaccessible by conventional endoscopy. Imaging modalities such as double-balloon enteroscopy, capsule endoscopy, CT angiography, intravenous contrast-enhanced multidetector row CT (MDCT) and magnetic resonance enterography (MRE) have been used to assist in the diagnosis of bleeding small intestine GISTs. The mainstay of management for small intestine GIST is complete surgical excision.

**CONCLUSION:** Bleeding jejunal GIST is very rare and only a handful of case reports have been published. The mainstay of management for small intestine GIST is complete surgical excision. It is essential to obtain a complete excision of localised disease and avoiding tumour spillage in order to reduce the risk of local recurrence and metastatic spread of GISTs.

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1. Introduction

Small intestinal bleeding is a rare presentation with a prevalence of 5–10% in patients presenting with gastrointestinal bleeding [6]. A patient presented with overt bleeding can be evaluated first with an upper and lower endoscopy to exclude upper and lower bleeding that can be readily reached with a standard endoscope [6]. GIST arising from the jejunum is a very rare cause of small intestinal bleeding [5]. The diagnosis of bleeding small intestine GISTs can be challenging as these are inaccessible by conventional endoscopy [4]. This case has been reported in line with the SCARE criteria [11].

2. Case presentation

A 66-year-old gentleman with no known comorbidities presented to us with a history of multiple melenic bowel movements. He has associated lethargy and easy fatigability as well. Further history from the patient revealed that he had been treated for symptomatic anaemia for the past two years. On clinical examination, he was pale, tachycardic but normotensive. His abdominal examination was unremarkable and digital rectal examination revealed melena. Initial investigations revealed a drop of haemoglobin from 11 g/dl to 4 g/dl. He was promptly resuscitated with blood products, and an early upper endoscopy was performed.

The index oesophagogastroduodenoscopy (OGDS) showed a small Forrest 3 antral ulcer with multiple subcentimeter gastric polyps [13]. The gastric polyps were biopsied, and the histopathology subsequently revealed it to be benign. A colonoscopy showed blood-stained colonic mucosa in its entirety. However, no bleeding source was identified. An urgent contrast-enhanced computed tomography (CECT) of the abdomen was performed which revealed...
no significant abnormality. There were no bowel related masses seen. After the acute gastrointestinal bleeding episode subsided, we proceeded to work him up with a presumptive diagnosis of possible small intestinal bleed.

Capsule endoscopy was performed which showed several small telangiectasias in the proximal part of the small bowel (Fig. 1). There was no visible tumour, polyps or ulcers. A double-balloon enteroscopy was then performed. It showed abnormal vascularity with a central umbilication over the mucosa of the small bowel

(Fig. 2). The mucosal abnormality was located at 165 cm from the incisor. Preoperative assessment of the patient showed he has an ASA score of 1 and a Revised Cardiac Risk Index score of 1 [12]. He subsequently underwent a laparoscopic examination in the theatre. Laparoscopic entry was performed with a closed technique (Veress needle) [14]. Intraoperative findings revealed an exophytic lesion measuring 6 cm x 6 cm x 3 cm approximately 30 cm distal to the duodenojejunal flexure (Figs. 3–5). Small bowel resection and primary side-to-side anastomosis were performed, without regional lymphadenectomy. He recovered uneventfully and was discharged after a short hospital stay.

The histopathological report confirmed the diagnosis of GIST arising from the jejunum, with a TNM staging of T3N0M0. The tumour measures 60 x 65 x 30 mm, with a mitosis of 1 per 50 high power field (HPF), with no evidence of tumour rupture. On immunohistochemistry staining, the tumour is positive for CD117 and DOG1. During his clinic follow up, he remains symptom-free
Small intestinal bleeding usually presents as stable overt or an occult bleed. A patient presented with haematemesis, melena or haematochezia is termed overt bleeding whereas an asymptomatic patient with faecal occult blood test positive is termed occult bleeding [6]. A patient presented with overt bleeding can be evaluated first with an upper and lower endoscopy to exclude upper and lower bleeding that can be readily reached with a standard endoscope. The terminal ileum should be intubated during colonoscopy to examine the ileal mucosa and to exclude bleeding source from a more proximal lesion [6]. Upper endoscopy in our patient revealed a small antral ulcer with multiple small gastric polyps without stigmata of recent haemorrhage whereas his colonoscopy does not reveal any significant abnormality.

Capsule endoscopy (CE) is the recommended first-line procedure for small bowel evaluation after upper and lower gastrointestinal bleeding source have been excluded [6]. It is usually performed before deep enteroscopy. Deep enteroscopy can be considered as the initial diagnostic tool if CE is contraindicated or in cases of massive haemorrhage [6]. When there is a strong suspicion of small bowel bleeding with abnormal VCE study, it should be followed by total deep enteroscopy [6]. Double-balloon enteroscopy is one of the methods of deep enteroscopy.

Gastrointestinal stromal tumours (GISTs) are the most common mesenchymal neoplasms of the alimentary tract but accounts for only 0.1–3% of all gastrointestinal neoplasms [1,2]. The most common sites of origin for GISTs are the stomach (60%–70%), the small intestine (25%–35%), the oesophagus (2%–3%), and rarely in the colon, the rectum or the appendix (5%) [3]. It is known to originate from the smooth muscle pacemaker interstitial cell of Cajal which has a function of coordinating gut motility and peristalsis [8].

The symptoms of GISTs are nonspecific and depend on the size and location. Small GISTs (<2 cm) are usually found incidentally from endoscopy or imaging for other pathology as many of them are asymptomatic. The most common symptom is gastrointestinal bleeding, which is present in 50% of the patients, followed by abdominal pain (20–50%) and gastrointestinal obstruction (10–30%) [3,8]. Other symptoms include melena, hematemesis, fullness, and a palpable mass. They rarely spread to the regional lymph node or other extra-abdominal organs, but they can metastasise to the liver [3,8].

According to the latest NCCN guidelines, a computed tomography (CT) scan of the abdomen is the initial workup for the evaluation, staging, and monitoring of treatment response. GIST on contrast-enhanced CT shows the characteristic of a well-defined soft tissue of relatively low density, which is homogenous [8]. In our patient, however, the CT of the abdomen failed to reveal any significant abnormalities. From the endoscopic view, GISTs form a well-delineated spherical or hemispheric mass, arising mostly from the muscularis propria layer beneath the mucosa and pushing it to the lumen to form a smoothly contoured elevation [8].

Surgical resection remains the mainstay treatment for non-metastatic GISTs [1,8]. Size of less than 2 cm with no signs of malignancy can be managed with active surveillance unless it is symptomatic then surgical resection is needed. However, a small tumour size does not exclude the malignant potential in a GIST [8]. Laparoscopic surgery has the advantage earlier recovery, less painful and has better cosmetic results. Furthermore, oncologic outcomes between laparoscopic and open surgery are similar [9,10]. As submucosal and lymphatic invasion is rare in GIST, this permits local excision rather than formal organ resection. Wide margins and lymph node dissections are not necessary. This allows laparoscopic resection to be one of the alternative techniques compared to more invasive open surgery [10].

The Asian consensus guidelines recommend laparoscopic resection for GIST up to 5 cm size when it is located in a favourable
4. Conclusion

GIST arising from the jejenum is a very rare cause of small intestinal bleeding. The diagnosis of bleeding small intestine GISTs can be challenging as these are inaccessible by conventional endoscopy. Normal findings from an upper and lower endoscopy in a patient presenting with gastrointestinal bleeding symptoms should raise a suspicion of small intestinal bleed. Imaging modalities such as double balloon enteroscopy, capsule endoscopy, CT angiography, intravenous contrast-enhanced multidetector row CT (MDCT) and magnetic resonance enterography (MRE) may be used to assist in the diagnosis of bleeding small intestine GISTs. The mainstay of management for small intestine GIST is complete surgical excision. It is essential to obtain a complete excision of localized disease and avoiding tumour spillage in order to reduce the risk of local recurrence and metastatic spread of GISTs.

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Ethical approval

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Consent

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Author contribution

Study concepts: Nik Ritza Kosai, Reynu Rajan. Study design: Nik Ritza Kosai, Reynu Rajan.

Data acquisition: Diana Mellisa Dualim, Loo Guo Hou. Quality control of data and algorithms: Nik Ritza Kosai, Reynu Rajan.

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