

Severe obscure gastrointestinal bleeding from Meckel's diverticulum with predominant ectopic pancreatic tissue

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Abstract

Obscure gastrointestinal bleeding represents about 5% of all gastrointestinal haemorrhages which is characterized by continuous or recurrent bleeding from an undetermined source after an initial bidirectional endoscopy. Meckel's diverticulum is a rare but recognized cause of obscure gastrointestinal bleeding. A carefully selected line of investigations is paramount to localize the causative lesion in obscure gastrointestinal bleeding which is a challenge in subacute cases. We present a case of 35-year-old female with thalassemia minor and mild anaemia presented with acute gastrointestinal bleeding from the ectopic pancreatic mucosa of an Meckel's diverticulum where only a small focus of gastric tissue was identified histologically during the follow-up. This case discusses the rarity of this histological presentation of Meckel's diverticulum as obscure gastrointestinal bleeding and the importance of intraoperative decision-making and intraoperative enteroscopy in cases of obscure gastrointestinal bleeding when other tests are negative.

Keywords

Meckel's diverticulum, ectopic pancreatic tissue, overt obscure gastrointestinal bleeding, diagnostic laparoscopy

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Introduction

Obscure gastrointestinal bleeding (OGIB) refers to persistent or recurrent bleeding that cannot be attributed to any conclusive source during the initial evaluation, which usually involves routine endoscopy.¹ The bleeding can be either visible (overt) or invisible (occult), depending on whether blood flow is apparent or not.² The small intestine is the most common location for OGIB. The causes of OGIB are vascular ectasia, small bowel tumours (gastrointestinal stromal tumours, adenocarcinomas, lymphoma, carcinoid and metastatic deposits), non-steroidal anti-inflammatory drugs or Crohn's induced ulcers and erosions. Aortoenteric fistula, Meckel's diverticulum (MD) and polyps are some of the other less-known causes of OGIB.³

MD is a common congenital abnormality of the gastrointestinal tract that affects approximately 2% of the general population. Most people with MD do not experience any symptoms, and it is often detected incidentally during surgery. However, in some cases, MD can cause serious complications such as intestinal obstruction, diverticulitis or gastrointestinal bleeding, which can significantly lower haemoglobin levels, particularly in adults.^{4–6} Other rare

complications include intussusception, hernia torsion, umbilical sinus or fistula and neoplasm.⁷ The incidence of complications is highest before the age of two and gradually decreases to almost none after 70 years.⁸ Incidence of MD-related complications is 1% after 40 years.⁸ Gastrointestinal bleeding is the most common presentation of MD.⁵ Ectopic glandular tissue in MD can be either gastric (~50%), endometrial, hepatobiliary, duodenal Brunner's glands and pancreatic mucosa or their combination.⁹

In practice, several techniques have been employed to identify MD with varied diagnostic efficacy such as Meckel's

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scan, small bowel capsule endoscopy (SBCE), balloon-assisted enteroscopy (BAE), computed tomography, angiography and small-bowel follow-through. The diagnostic accuracy of BAE is much higher than that of Meckel's scan.¹⁰ When there is an overt bleeding, an arteriography can be used to determine the cause of the bleeding.⁸

Here, we report a case of OGIB in a 35-year-old female caused by MD, which was discovered during an exploratory laparotomy after a series of negative diagnostic investigations and a rare histology of predominant ectopic pancreatic tissue with a single focus of gastric tissue was found within the MD.

Case presentation

A 35-year-old Sri Lankan female presented with a history of recurrent fresh per rectal bleeding for past 7 years. She had symptoms of anaemia on admission with a haemoglobin of 5 g/dl. Three previous similar episodes had spontaneously resolved and extensively investigated without an obvious cause. Each of those episodes was managed with blood transfusions. She was diagnosed with thalassemia minor and mild anaemia. On examination, the patient was pale, tachycardia, had a weak pulse systolic blood pressure was 85 mmHg. A digital rectal examination revealed altered blood with clots.

Following resuscitation and stabilization, a computer tomographic mesenteric angiogram (CTA), upper (up to proximal jejunum) and lower gastrointestinal endoscopy and ileal intubation were performed. CTA was negative for an active bleeding site, and the upper gastrointestinal endoscopy including a limited endoscopy was normal. But the colonoscopy revealed altered blood in the caecum and ascending colon, but did not show an active bleeding site in the colon.

In addition, a repeat SBCE showed evidence of fresh blood in the distal ileum and ascending colon. The impression was that the bleeding originated from the colon rather than the ileum.

Her contrast computed tomography was unremarkable. While in the ward, she developed a massive per rectal bleeding. As she was hemodynamically unstable and rest of the focused investigations didn't reveal a cause for the bleeding, a diagnostic laparoscopy was planned in the middle of the night after resuscitation. Intra-operatively, we found that some of the proximal small intestine loops and the ascending colon were filled with blood, and there was a MD (Figure 1) with thickened mucosa at the tip. Repeated examination of the rest of the small bowel was normal from the duodenojejunal flexure to the ileocecal junction. An intra-operative decision was made to perform an intra-operative endoscopy via an appendostomy was performed to exclude any right colon pathology which was suspected in the SBCE. As there was no access to an endoscopy due to practical reasons, had to improvised and used a mobile video bronchoscope available in the ICU to

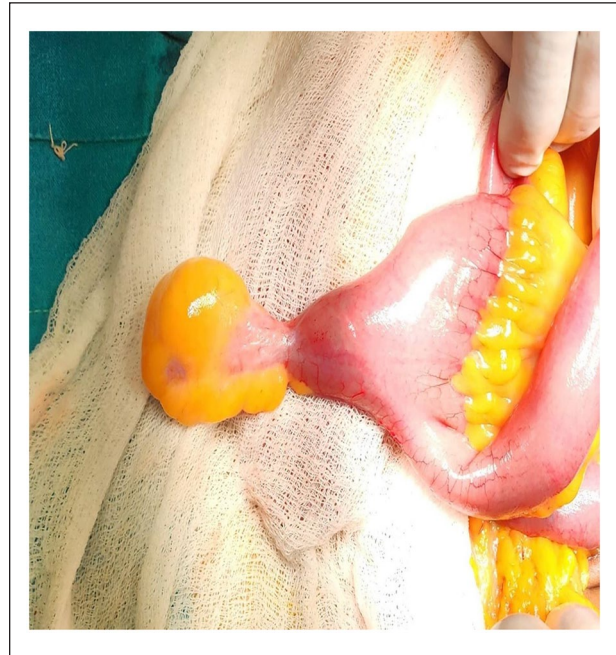


Figure 1. Intraoperative image of the Meckel's diverticulum.

intra-operatively see the bowel for a bleeding site. The video bronchoscope is a 6mm diameter and approximately 25 cm length scope without ability to inflate air but able to suck minimal secretions. As we had to see both the colon and distal ileum before performing a resection and scope was short with minimal functions, then decided to enter through the appendix which will not cause any unnecessary damage to the bowels. Fortunately, there was minimal blood in the ascending colon, which confirmed that the bleeding was unlikely from the colon and this made us more confident in to perform Wedge resection of MD and appendectomy (Figure 2).

She had an uneventful post-operative period and was discharged with analgesics in a couple of days. Histopathology revealed a MD with predominant pancreatic mucosa and a focus of gastric mucosa (Figure 3). The patient was well with haemoglobin of 10.7 g/dL in her 1-month review.

Discussion

MD and its complications, particularly gastrointestinal bleeding is more common in children than adults.⁵ Moreover, diagnosis of MD is overlooked in adults, and there are no predictive factors to determine the occurrence of complications. But the risk of complications reduces as the patient ages.⁵

Yamaguchi et al evaluated 600 cases of pre-operatively suspected MD, out of which only 34 were actually diagnosed with MD, resulting in a diagnostic rate of 5.7%. The most common complication of this illness was intestinal obstruction, followed by intussusception, inflammation and haemorrhage in that order in his series.¹¹ Out of the 93 cases of MD with ectopic tissue, 58 had ectopic stomach mucosa.¹¹

Ectopic tissue is often associated with symptomatic Meckel's disease, particularly with gastrointestinal bleeding.¹² Ectopic tissue can also be present in silent MD, although it is less common. Ectopic gastric tissue is found in 0%–18.2% of cases, while ectopic pancreatic tissue is present in 0%–5.0% of cases.¹² According to a systematic

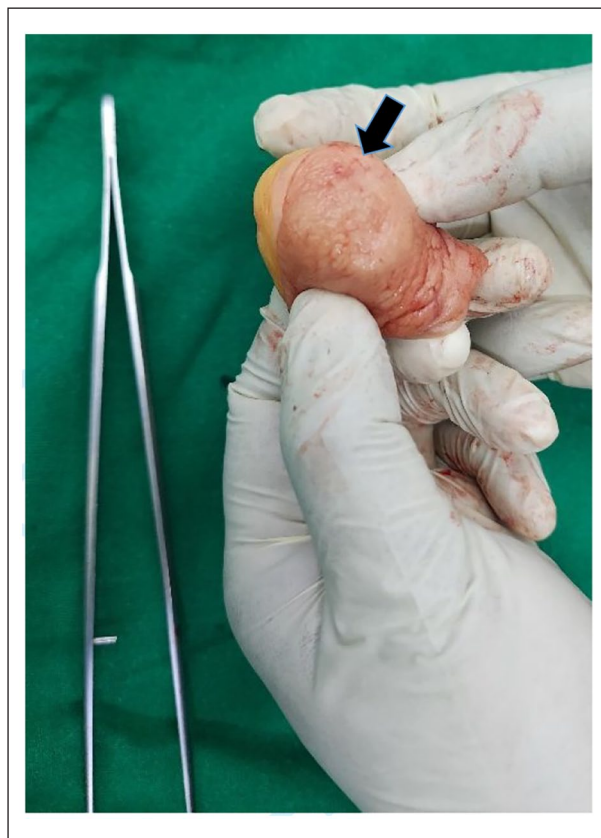


Figure 2. Opened resected specimen of Meckel's diverticulum: Thick arrow shows the suspected area of an ulcer.

review, ectopic pancreas and stomach tissue account for 97.0% of all ectopic tissues. Ectopic duodenal and colonic tissues are less common.¹²

Almost all patients with bleeding MD have heterotopic mucosa, of primarily gastric origin.⁵ Autopsy studies have revealed that the heterotopic pancreas (HP) affects a low percentage of the population, ranging from 0.5% to 14.0%.¹³ This clinical condition mostly affects the stomach, duodenum and jejunum, but it can also occur in any site of the digestive system, including the oesophagus, terminal ileum, omentum, mesentery, spleen, liver, MD, colon, gallbladder, urinary bladder, lung and lymph nodes.¹³ Compared to the ileum, the proximal small intestine, duodenum and stomach are more likely to have heterotopic pancreatic tissues that occurs alone. This condition is frequently asymptomatic.¹⁴

Jean-Schultz originally described HP in 1729. HP is defined as pancreatic tissue without a true anatomical or circulatory link to the pancreas.¹⁵ Inflammation, ulceration, chemical irritation, haemorrhage, blockage, malignant transformation, jejunojejunal intussusception and ileus are among the problems that HP may induce.^{16–19} Nevertheless, the majority of ectopic pancreatic patients do not exhibit any symptoms, and the diagnosis is typically verified by radiography, endoscopy or surgical investigations driven by other medical conditions.²⁰

It is challenging to diagnose HP in the small intestine before surgery. The extent of the lesion and the mucosal involvement determine the symptoms. Significant lesions are larger than 1.5 cm on a clinical examination.^{18,20} The concomitant finding of ectopic pancreatic tissue along with gastric tissue in MD is never reported in the literature in any age group. There are only a few reports of ectopic and heterotopic pancreatic tissue in MD in children²¹ and a single report of heterotopic pancreatic tissue in adults which presented with overt OGIB.⁹

Gastrointestinal bleeding from MD is generally caused by acid production from an ectopic mucosa, particularly from

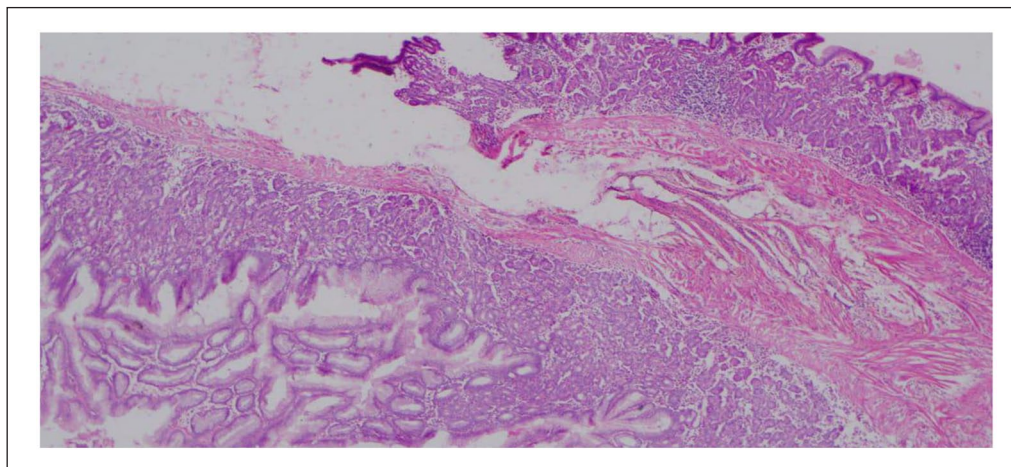


Figure 3. The histological assessment showed a single gastric tissues focus with the majority of ectopic pancreatic tissue.

the stomach, acid-induced ulceration of the adjacent ileal mucosa. More rarely, bleeding can be a result of recurrent diverticular intussusception which causes recurrent erosion of the ileal mucosa.⁵ However, MD-related bleeding is exceedingly rare in adults and has only been reported in case studies in the English literature.²²

Pathological examination is the most reliable diagnostic technique for identifying ectopic pancreas. However, most biopsies are unsatisfactory as it is difficult to obtain sufficient tissue samples.²³ The 99m technetium pertechnetate scintigraphy is a method that can be used to identify ectopic gastric mucosa in MD. However, its diagnostic accuracy in adults is only 46%.¹⁷ X-rays, CT scans and abdominal ultrasonography often produce vague results.²³ Double-balloon enteroscopy or capsule endoscopy can be used to identify MD. However, it is challenging to diagnose MD with HP.^{17,23}

In our patient, the diagnosis of MD was not picked up since 2017 with the background of severe life-threatening gastrointestinal bleeding which caused significant morbidity. In this patient a diagnostic laparoscopy with or without intra-operative enteroscopy would have been the ideal option from the outset when the other focused investigations did not reveal a cause. Ba et al reported 22 cases of massive OGIB [5 were due to MD], where they successfully identified the source of bleeding by laparoscopy and laparoscopy-assisted enterectomy, and enteroanastomosis was performed in most of those cases.²⁴

Resection of the unintentionally found MD is justified in males, lengthy diverticula (greater than 2 cm), the presence of ectopic tissue and patients under the age of 50 years.⁸ The best treatment for symptomatic MD is surgical excision.⁸ Diverticulectomy, wedge resection and segmental resection are recognized surgical techniques, and the choice usually depends on the existence and location of ectopic tissue, as well as the integrity of the diverticular base and surrounding ileum.⁸

Conclusion

MD is recognized but as an uncommon cause of OGIB. While MD can be asymptomatic or present with complications, bleeding MD with predominant ectopic pancreatic tissue is especially rare. Accurate diagnostic techniques are crucial for patients with OGIB to identify the causative lesion, particularly in subacute cases. When initial diagnostic techniques fail, diagnostic laparoscopy and intraoperative enteroscopy should be considered.

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Author contributions

D.S design the manuscript. A.J. Writing the manuscript and assisting surgeon of the case; O.M.O. Revision of the manuscript,

assisting surgeon of the case; G.R. Histopathological analysis of the case and providing images of histology slides; N.F. Intra-operative endoscopist and revision of the manuscript; D.S. Supervisor of the case, primary surgeon of the case and revision of the manuscript.

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Ethics approval

Our institution does not require ethical approval for reporting individual cases or case series.



Informed consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Research registration

None.

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