

## Inverted Meckel's diverticulum: Two case reports and a review of the literature

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### Abstract

Gastrointestinal surgeons seldom encounter inverted Meckel's diverticulum in their clinical practice. We describe two cases of inverted Meckel's diverticulum. If the patient has a disease-related complication such as intussusception, as with our first case, it can be easily detected. However, if the patient has subacute or chronic symptoms, as with our second case, the diagnosis might be delayed. Regardless of the disease-related complication, intussusception of inverted Meckel's diverticulum can be easily managed with laparoscopic single-port surgery.

**Key words:** Inverted Meckel's diverticulum; Laparoscopic surgery; Intermittent hematochezia; Intussusception; Abdominal pain

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**Core tip:** When clinicians encounter an adult patient complaining of intermittent hematochezia and/or abdominal pain without any evidence of gastrointestinal bleeding after esophagogastroduodenoscopy and co-

lonoscopy, inverted Meckel's diverticulum and other small bowel pathologies must be considered to avoid unwanted complications related to these rare disease entities. Computed tomography scan is a beneficial diagnostic tool to identify small bowel pathology and to differentiate among diverse diseases, including lipomas, inflammatory fibroid polyps, vascular malformations, lymphomas, inverted diverticula and malignant tumors.

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## INTRODUCTION

Meckel's diverticulum is a congenital anomaly that results from persistence of the omphalomesenteric duct at its attachment site to the distal ileum. According to autopsy studies, this condition is found in 2% of the general population<sup>[1,2]</sup>. On average, the diverticulum is located two feet proximal to the ileocecal valve and often contains gastric or pancreatic tissue, causing mucosal bleeding. Patients with Meckel's diverticulum are usually asymptomatic. However, according to a population-based study, untreated Meckel's diverticulum cases are associated with as high as a 6.4% lifetime risk of developing complications requiring surgical treatment<sup>[3]</sup>. The most common presentation of complicated Meckel's diverticulum is gastrointestinal (GI) bleeding; less frequent symptoms include diverticulitis, volvulus and intestinal obstruction with or without intussusception, a condition in which a proximal segment of bowel rolls into a distal part<sup>[4]</sup>. To the best of our knowledge, inversion of a Meckel's diverticulum is rare and has been reported in only a few case series worldwide<sup>[5-8]</sup>.

## CASE REPORT

### Case 1

A 34-year-old man without any underlying disease visited our gastroenterology clinic on October 29, 2014 for melena beginning 5 d earlier. The initial laboratory findings were all within normal ranges, and subsequent esophagogastroduodenoscopy showed no remarkable findings except for several areas of erosive gastritis. The patient was diagnosed with irritable bowel disease, but the symptoms remained intractable after one year of follow-up. On March 16, 2016, the patient complained of hematochezia with left lower quadrant abdominal pain. Emergent sigmoidoscopy was performed but failed to reveal any focus of bleeding. Thereafter, computed tomography (CT) scans revealed small bowel intussusception with segmental thickening of the mucosa (Figure 1A). The patient was referred to the Depart-

ment of General Surgery, and laparoscopic single-port exploration was performed. During the operation, bowel edema at the distal ileum (50-60 cm upstream of the ileocecal valve) was observed, and an intraluminal mass was palpable. Securing the margin, segmental resection of the small bowel with side-to-side stapled anastomosis was performed. The delivered specimen was inspected, and the intraluminal polyp-like mass was exposed (Figure 1B). The patient had an uneventful postoperative course and was discharged 6 d after surgery. Final pathology revealed an inverted intussusception of Meckel's diverticulum with focal heterotopic gastric mucosa (Figure 1C).

### Case 2

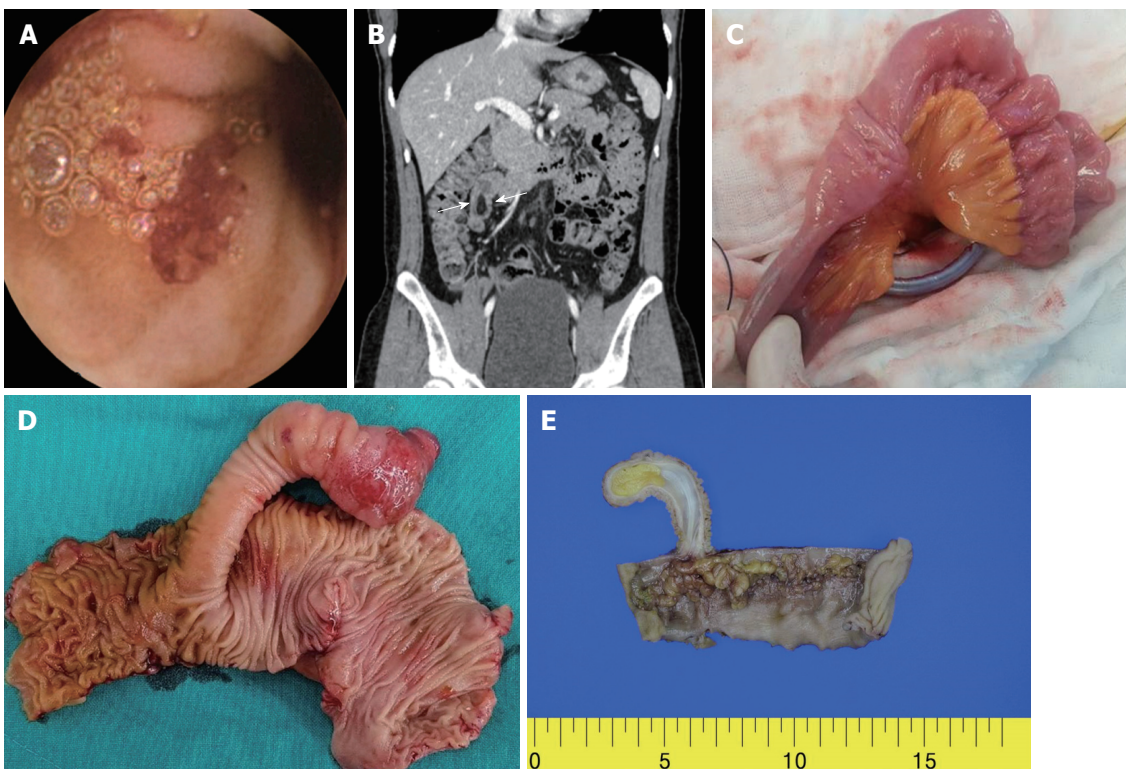
A 27-year-old man with a history of heavy alcohol consumption visited the emergency room on December 31, 2017 for hematochezia beginning 4 d earlier. The initial laboratory findings showed low hemoglobin levels (8.5 g/dL). Serial physical examinations revealed old clots on irrigation through a Levin tube and melena upon digital rectal examination. Upon admission, two pints of packed red blood cells were transfused, and emergent esophagogastroduodenoscopy with sigmoidoscopy failed to reveal intestinal pathology. Therefore, the gastroenterologist decided to perform capsule endoscopy. This procedure revealed small bowel bleeding, but failed to detect intestinal pathology due to low video quality (Figure 2A). A CT scan revealed small bowel intussusception at the distal ileum with a possible 2 cm lipoma (Figure 2B). After a diagnosis of intussusception, the patient was referred to the Department of General Surgery. On January 4, 2018, laparoscopic single-port exploration was performed. During surgery, the intussusception at the distal ileum with a palpable club-like intraluminal mass was detected (Figure 2C), and segmental small bowel resection with end-to-end anastomosis was performed. While exploring the specimen, a large polyp-like mass with mucosal ulceration at the tip was demonstrated in the small bowel (length 6 cm, width 1.5 cm). The patient had an uneventful postoperative course and was discharged 6 d after surgery. The final pathology revealed that the mucosa showed a finger-like projection with normal mucosa and a surface ulceration measuring 5 cm × 2 cm × 2 cm (Figure 2D). Upon sectioning of the finger projection, the cut surface showed serosa infolding with mesenteric tissue, consistent with an inverted diverticulum causing intussusception without heterotopic tissue (Figure 2E).

## DISCUSSION

The mechanism of inversion in Meckel's diverticulum is still not clearly understood. One possible explanation is that abnormal peristaltic movement around an ulceration or ectopic tissue at Meckel's diverticulum may cause inversion<sup>[9]</sup>. Inverted Meckel's diverticulum can cause two clinical manifestations: GI bleeding and intussusception,



**Figure 1** Inverted Meckel's diverticulum causing intussusception with focal heterotopic gastric mucosa. A: Computed tomography scan: small bowel intussusception with segmental thickening of mucosa at the distal ileum; B: Bowel edema at the distal ileum (50-60 cm upstream from ileocecal valve) with a polyp-like mass; C: Inverted intussusception of Meckel's diverticulum with focal heterotopic gastric mucosa. White arrow: typical target sign; yellow circle: goblet cells; yellow arrow: pyloric glands; red arrow: foveolar epithelium; blue arrow: small intestinal epithelium.



**Figure 2** Inverted Meckel's diverticulum causing intussusception without heterotopic tissue. A: Capsule endoscopy: small bowel bleeding, but failed to detect intestinal pathology; B: Computed tomography scan: small bowel intussusception at the distal ileum with possible 2 cm sized low-density mass (white arrow); C: Laparoscopic finding: the intussusception at the distal ileum with palpable club-like intraluminal mass; D: Finger-like projection with normal mucosa and surface ulceration; E: Section of finger-like projection: serosa in folding with mesenteric tissue, consistent with inverted diverticulum causing intussusception without heterotopic tissue.

as in our two cases. Most complaints are of hematochezia and/or melena due to ulceration of the inverted Meckel's diverticulum. Ulceration may be caused by ectopic gastric or pancreatic tissues in the inverted Meckel's diverticulum itself, but can also occur without accompanying abnormal ectopic tissues. The latter can be explained by repetitive mechanical trauma to the mucosa from the reversible intussusception.

CT is a common tool for diagnosing intussusception in the small intestine. A characteristic coiled spring appearance or target sign are well-known findings on CT scans. However, if the patient visits the clinic complaining

of melena or hematochezia without abdominal pain, as in Case 2, the diagnosis may be delayed. Pantongrag-Brown *et al*<sup>[8]</sup> reported that approximately 80% of patients with inverted Meckel's diverticulum complained of subacute or chronic symptoms, including GI bleeding, intermittent abdominal pain, or other signs of low-grade small bowel obstruction<sup>[8]</sup>. Although capsule endoscopy can help confirm small bowel bleeding, as in Case 2, routine use of this device for diagnosing small bowel bleeding must be done with caution. This caution is necessary not only due to low video quality and its limitations regarding exact location of the intestinal pathology<sup>[10]</sup>, but also

because of the increased risk of intestinal obstruction. On the other hand, CT scans are much more beneficial for the identification of small bowel pathology and differentiation of diverse diseases, including lipomas, inflammatory fibroid polyps, vascular malformations, lymphomas, inverted diverticula and malignant tumors. Inverted Meckel's diverticulum is sometimes confused with lipoma on CT scans. However, findings of an intraluminal polypoid lesion in the small intestine covered with a thick collar of enhancing soft tissue may be a decisive indicator of inverted Meckel's diverticulum as opposed to lipoma, which appears on CT scans only as a thin covering over the low-density fatty mass<sup>[5]</sup>.

Intermittent and/or small amounts of GI bleeding can be easily detected by Tc<sup>99m</sup> RBC scintigraphy, where the minimum detectable bleeding rate was reported as 0.05-0.2 mL/min; relatively stable persistence in the circulation made it possible to monitor patients with intermittent bleeding<sup>[11]</sup>. Scintigraphy could have been used to locate the bleeding focus in the first case. However, according to the gastroenterologist, they initially thought that the chronic intermittent hematochezia in this patient was due to a hemorrhoid. This was why the patient was followed by a local gastroenterology clinic for one year. Subsequently, the patient was admitted to the emergency room complaining of hematochezia with abdominal pain. CT scans were used to evaluate the cause of the left lower quadrant pain but not the hematochezia in this case. In the second case, scintigraphy could have been used to evaluate the cause of melena. However, the gastroenterologist decided on capsule endoscopy, revealing GI bleeding in the small intestine. According to the gastroenterologist, scintigraphy was not considered at that moment because the bleeding from the small intestine was already confirmed by capsule endoscopy and because CT scans are performed to reveal underlying pathologies causing GI bleeding in adults. CT angiography can be considered if the GI bleeding is active. The detection limit of CT angiography for active bleeding was 0.3 mL/min in porcine models<sup>[11]</sup>. Our cases were regarded as having chronic and intermittent GI bleeding, not active bleeding.

Although the gastroenterologist had 30 years of experience, they performed capsule endoscopy in the second case. They expected to find small bowel pathology but failed to do so. Apart from low video quality, if the capsule was obliterated by small bowel pathologies, the patient might experience intestinal obstruction, leading to increased morbidity related to surgery. Following this report, we hope clinicians worldwide will be cautious regarding the use of capsule endoscopy to evaluate the cause of GI bleeding in adults.

Currently, small bowel intussusception in adults, regardless of cause, requires prompt surgical management, such as segmental resection of the small bowel. A thorough appreciation of the characteristic CT scan findings of inverted diverticulum (as opposed to those of malignant tumors such as lymphoma) may help create

an appropriate surgical approach.

## ARTICLE HIGHLIGHTS

### Case characteristics

The chief complaint of adults with inverted Meckel's diverticulum is intermittent hematochezia, melena and/or abdominal colicky pain.

### Clinical diagnosis

If an adult patient has chronic intermittent abdominal pain, melena, hematochezia, and/or complaints of intestinal obstruction, inverted Meckel's diverticulum with intussusception should be considered.

### Differential diagnosis

CT scans would be beneficial for differentiating among other diseases such as lipoma, inflammatory fibroid polyps, vascular malformations, lymphoma, and malignant tumors.

### Laboratory diagnosis

In cases of intermittent hematochezia and/or melena, hemoglobin level would reveal anemia.

### Imaging diagnosis

A characteristic coiled spring appearance or target sign are well known findings on CT scans.

### Pathological diagnosis

Full-thickness inversion of diverticulum with or without heterotopic tissues such as stomach, pancreas *etc.* in hematoxylin and eosin staining would be the main finding of inverted Meckel's diverticulum.

### Treatment

Small bowel intussusception in adults, regardless of cause, requires prompt surgical management, such as segmental resection of the small bowel.

### Related reports

Meckel's diverticulum is a congenital anomaly of the omphalomesenteric duct remnant attached to the distal ileum and, when left untreated, the lifetime risk of developing complications requiring surgical treatment can be as high as 6.4%

### Term explanation

Intussusception means a condition in which a proximal segment of bowel rolls into a distal part and causes intestinal obstruction.

### Experiences and lessons

Following this report, we hope clinicians worldwide will be cautious regarding the use of capsule endoscopy to evaluate the cause of gastrointestinal bleeding in adults.

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