

Inverted Meckel's Diverticulum Masquerading as a Subepithelial Tumor of the Ileum

Mohammad Noubani, BS¹, Rohit Bhan, MS¹, Nimra Ghani, BS¹, Pons Materum, MD², Emily Glazer, MD³, Georgios Georgakis, MD, PhD¹, and Jonathan M. Buscaglia, MD³

¹Department of Surgery, Renaissance School of Medicine, Stony Brook, NY

²Department of Pathology, Renaissance School of Medicine, Stony Brook, NY

³Division of Gastroenterology, Department of Medicine, Stony Brook, NY

ABSTRACT

We describe a case of an inverted Meckel's diverticulum presenting as an intraluminal subepithelial lesion on intraoperative enteroscopy. A 53-year-old woman presented with chronic iron deficiency anemia unresponsive to escalating iron supplementation. After equivocal upper and lower endoscopy and negative cross-sectional imaging, capsule endoscopy suggested a submucosal mass lesion in the proximal ileum. Antegrade double-balloon enteroscopy was unsuccessful in reaching the lesion. A large pedunculated, submucosal-appearing lesion was finally identified on intraoperative enteroscopy. The mass was surgically resected, and final pathology confirmed an inverted Meckel's diverticulum.

INTRODUCTION

Inverted Meckel's diverticula are very rare causes for unexplained occult gastrointestinal (GI) bleeding.¹ Although some sources report that up to 21% of Meckel's diverticula are inverted, only 40 case reports have described this disease as a cause of intussusception or occult bleeding.² We describe a case report for a Meckel's diverticulum identified on intraoperative enteroscopy after a failed attempt at double-balloon enteroscopy (DBE).

CASE REPORT

A 53-year-old woman presented with no overt GI bleeding and recurrent iron deficiency anemia unresponsive to oral iron supplementation. Her most recent Hgb/Hct before endoscopy was 9.6 g/dl/30.8, and her iron studies showed an iron saturation of



Figure 1. Wireless capsule endoscopy showing a prominent fold in the proximal ileum suspicious for a subepithelial mass lesion. Arrow here is showing a prominent fold in the proximal ileum suspicious for a subepithelial mass lesion.

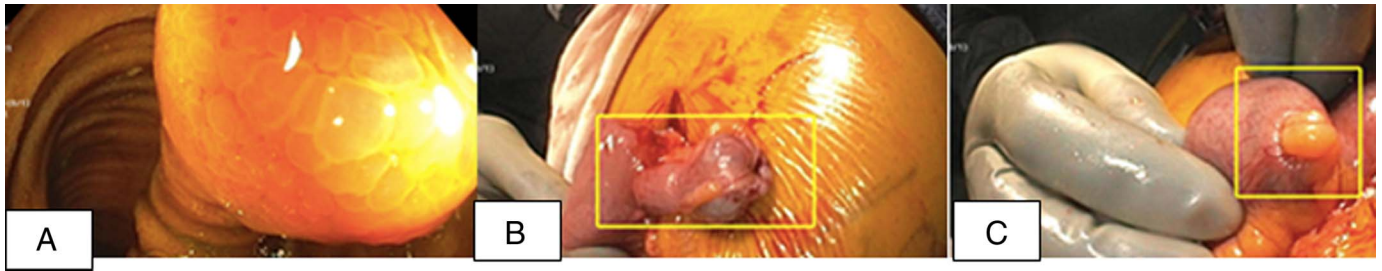


Figure 2. Endoscopy of the diverticulum showing (A) an endoscopic polyp in the lumen of the ileum, (B) an inverted diverticulum pulled from the laparotomy incision, and (C) the opening of the diverticulum.

12% and a total ferritin of 27 $\mu\text{g/L}$. She underwent a routine GI evaluation with unremarkable findings on upper endoscopy and colonoscopy with terminal ileoscopy. A computed tomography enterography with IV contrast yielded a normal small intestine. Owing to persistent anemia with heme-positive stool, a wireless capsule endoscopy was performed to further examine the small bowel. The wireless capsule endoscopy study revealed a prominent fold in the proximal ileum, suspicious for a subepithelial mass lesion (Figure 1). An antegrade DBE procedure was attempted yet technically challenging because of adhesions of no clear etiology given the negative surgical history of the patient. The point of maximal insertion of the enteroscope within the jejunum was marked with a tattoo. Given the equivocal workup for this patient's chronic blood loss and absolute diagnoses and treatment of her underlying mass lesion, a decision was made to perform intraoperative push enteroscopy in an effort to further evaluate the small bowel distal to the tattoo mark.

After laparoscopic exploration of the peritoneal cavity, a small enterotomy was created in the mid jejunum. A standard pediatric colonoscope was then inserted through the enterotomy and advanced in an antegrade fashion beyond the tattoo and into the distal small intestine. Within the proximal ileum, a large 3-cm pedunculated, polypoid lesion was readily identified

(Figure 2). The ileal segment containing the mass was retrieved from the laparotomy incision (Figure 2). The mass was reduced resembling a Meckel's diverticulum (Figure 2). The identified lesion was resected using a stapler device without any disturbance of the involved ileal segment. The specimen was collected and sent to pathology for final diagnosis. Pathology results confirmed an inverted Meckel's diverticulum with mucosal ulceration, granulation tissue, and foreign-body giant cell reaction (Figure 3). The resection margins were unremarkable containing the normal small bowel mucosa and wall.

DISCUSSION

Meckel's diverticula are common causes of occult GI bleeding/iron deficiency anemia in childhood.¹ They are much less common in adulthood. Inverted Meckel's diverticula are extremely rare and difficult to diagnose. To the best of our knowledge, this is the first case report of an inverted Meckel's diverticulum to be diagnosed on intraoperative enteroscopy after a failed DBE, despite the increased use of DBE for the diagnosis of Meckel's diverticula.³

This case was difficult to diagnose with conservative management, particularly because of the lack of symptoms, normal

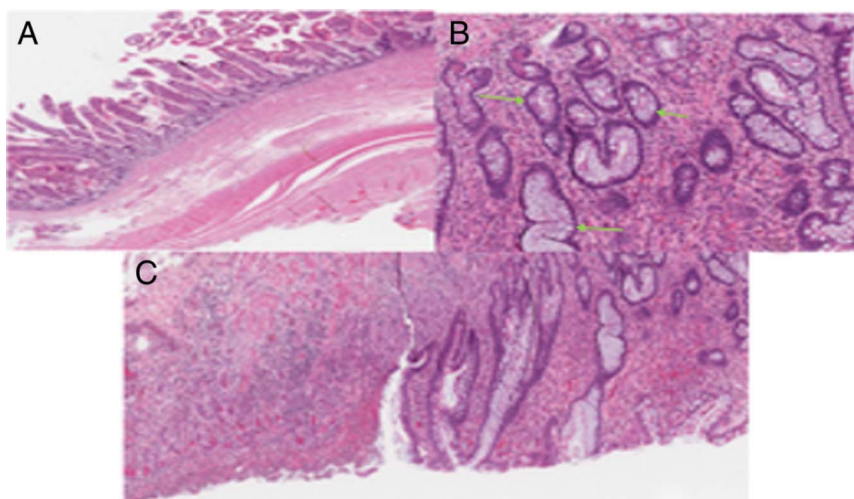


Figure 3. Histology of the Meckel's diverticulum showing (A) the normal intestinal mucosa, (B) pyloric metaplasia of the diverticulum, and (C) mucosal ulceration with granulation tissue on the tip of the polypoid lesion. Arrows in here are showing pyloric metaplasia.

endoscopic findings, and unremarkable imaging. With this patient's occult blood loss and equivocal computed tomography enterography, it was challenging to identify the source of bleeding. Furthermore, advanced imaging such as mesenteric angiography and radioisotope-labeled red blood cell scintigraphy would not have been viable given the absence of active bleeding in this patient.⁴ Therefore, after an unsuccessful attempt at per os enteroscopy with antegrade DBE, an intraoperative push enteroscopy was recommended to better evaluate the cause of occult bleeding in this patient.⁵

The etiology behind the Meckel's diverticulum inversion is controversial, with some sources citing as many as 21% of the diverticula being inverted.² Some evidence suggests that the lack of Meckel's mesenteric fixation permits inversion.⁶ Furthermore, small bowel peristalsis near the Meckel's diverticulum could contribute to inversion.⁷ Through this inversion, the Meckel's diverticulum could cause problems beyond occult bleeding and iron deficiency anemia from the heterotopic gastric mucosa. It can act as a pathological lead point for intestinal intussusception and small bowel obstruction as noted in several other previous reports.⁸

This case presents an inverted Meckel's diverticulum identified with intraoperative push enteroscopy and confirmed on pathology findings of the heterotopic gastric mucosa. Despite the rarity of this disease, an inverted Meckel's diverticulum should be on the differential diagnosis for occult GI bleeding in the setting of unremarkable imaging and uncertain endoscopy findings. Intraoperative push enteroscopy is a safe and viable option for patients with a failed DBE procedure.

DISCLOSURES

Author contributions: M. Noubani wrote the manuscript, reviewed the literature, and is the article guarantor. R. Bhan and N. Ghani wrote the manuscript, reviewed the literature, and provided the endoscopic images. P. Materum wrote the manuscript, reviewed the literature, and provided the pathology

images. E. Glazer edited the manuscript and provided the endoscopic images. G. Georgakis wrote the manuscript and revised the manuscript for intellectual content. J. Buscaglia wrote the manuscript, reviewed the literature, provided the endoscopic images, and revised the manuscript for intellectual content.

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