

## Inverted Meckel's diverticulum as a cause of occult lower gastrointestinal hemorrhage

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### Abstract

Meckel's diverticulum is a common asymptomatic congenital gastrointestinal anomaly, but rarely it can present with hemorrhage. Over the last few years inverted Meckel's diverticulum has been reported in the literature with increasing frequency as an occult source of lower gastrointestinal hemorrhage. Here, we report a case of a 54-year-old male, who was referred for surgical evaluation with persistent anemia and occult blood per rectum after a work up which failed to localize the source over 12 mo, including upper and capsule endoscopy, colonoscopy, enteroclysis, Meckel scan, and tagged nuclear red blood cell scan. An abdominal computed tomography scan showed a possible mid-ileal intussusception and intraluminal mass. During the abdominal exploration, inverted Meckel's diverticulum was diagnosed and resected. We review the literature, discuss the forms in which the disease presents, the diagnostic

modalities utilized, pathological findings, and treatment. Although less than 40 cases have been reported in the English literature from 1978 to 2005, 19 cases have been reported in the last 6 years alone (2006-2012) due to improved diagnostic modalities. Successful diagnosis and treatment of this disease requires a high index of clinical suspicion, which is becoming increasingly relevant to general gastroenterologists.

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**Key words:** Inverted Meckel's diverticulum; Gastrointestinal hemorrhage; Lower gastrointestinal bleeding; Intussusceptions

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### INTRODUCTION

Meckel's diverticulum is the most common congenital abnormality in the gastrointestinal tract, but is usually asymptomatic<sup>[1]</sup>. When symptomatic it may present with hemorrhage in association with ectopic gastric and/or pancreatic mucosa, intestinal obstruction, intussusceptions, or inflammation. This abnormality can also present in the setting of an inverted diverticulum causing a lower gastrointestinal bleed. There is no role for non-operative

management in inverted Meckel's diverticulum, which mandates early surgical removal. Here, we present a case which first underwent an extensive diagnostic evaluation of persistent gastrointestinal hemorrhage over a 12-mo period before this entity was diagnosed and then appropriately treated. The aim of this report is to alert surgeons and gastroenterologists of this important source of persistent gastrointestinal hemorrhage which requires a high clinical suspicion to diagnose because of how difficult it is to detect. We conducted an extensive review of the previously reported cases in the literature, and discuss the presentations of this readily curable disease, the utility of various diagnostic modalities, pathological findings, and the appropriate management.

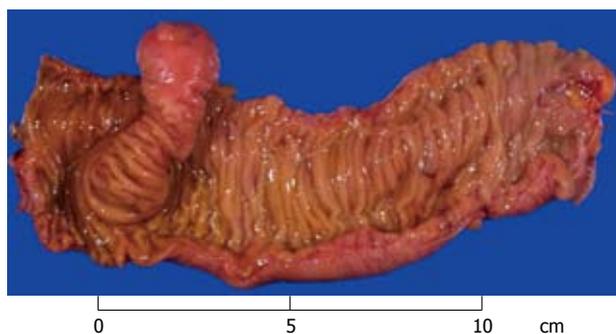
## CASE REPORT

A 54-year-old male with no significant past medical history was seen by a gastroenterologist for anemia and stools positive for occult blood. An extensive diagnostic evaluation was undertaken over a 12-mo period before the patient was referred for surgical evaluation. He had a negative upper gastrointestinal and capsule endoscopy, and a negative colonoscopy, despite adequate bowel preparation. Enteroclysis revealed a polypoid lesion in the mid jejunum. The differential diagnosis at that point included tumor, lipoma, carcinoid, or sarcoma. He underwent a Meckel scan, which was negative. Given the continued anemia and occasional bright red blood per rectum, he underwent a tagged nuclear red blood cell scan, which failed to demonstrate an acute hemorrhagic source. An abdominal computed tomography (CT) scan was obtained, which demonstrated a possible mid-ileal intussusception and an intraluminal mass. The patient was referred for a surgical evaluation.

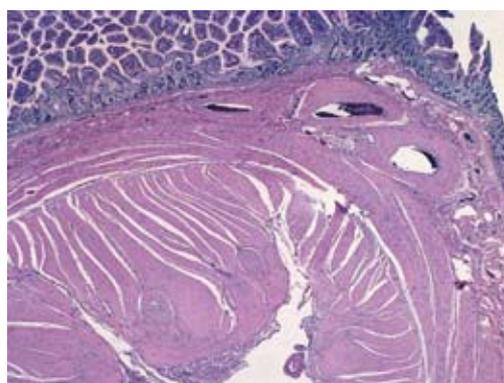
The patient underwent an exploratory laparoscopy which demonstrated a hard intraluminal mass in the mid-ileum. The remainder of the small bowel was normal to the level of the ligament of Treitz. A 5 cm mini midline-laparotomy was performed overlying the small bowel lesion. The lesion containing portion of the small bowel was delivered out of the abdomen and resected. Small bowel continuity was established *via* a stapled side-to-side anastomosis.

On gross examination, the specimen consisted of a segmental resection of the small bowel with a small dimple shaped defect penetrating through the bowel wall. This defect was proven to be patent by a probe. Opening of the specimen revealed a 5.0 cm × 1.8 cm × 1.5 cm polypoid lesion that terminated in a club-shaped head (Figure 1). The small bowel mucosa was contiguous with that of the stalk and extended to within 1.5 cm of the tip of the lesion. The dumbbell shaped tip was covered by attenuated mucosa that was granular in appearance. A small area of erythema was seen at the tip and appeared grossly consistent with an area of hemorrhage.

Microscopic sections revealed well-defined intestinal mucosa that surrounded the core of the lesion (Figure 2). The most internal component of the core consisted of



**Figure 1** Gross specimen of the inverted Meckel's diverticulum arising in the segmental resection of the small bowel.



**Figure 2** Histopathology reveals a central core consisting of serosa and muscle. The cross section reveals small intestinal mucosa lining the fragment.

the serosa and the muscle of the bowel wall. These layers were circumscribed by the intestinal submucosa and mucosa that became attenuated toward the tip of the specimen. The mucosa seen at the tip was that of intestinal-type with scattered paneth cells (exocrine serous cells) and goblet cells. No heterotopic gastric or pancreatic tissue was identified. The patient had an uneventful post-operative recovery. He was discharged on post-operative day three, and never had further episode of lower gastrointestinal hemorrhage nor any complications or sequelae during the subsequent two years of follow up.

## DISCUSSION

Meckel's diverticulum is the most common congenital anomaly in the gastrointestinal tract<sup>[1]</sup>. In an autopsy series, the incidence was reported as 1%-3%<sup>[2]</sup>. Johann F Meckel, a famous anatomist, was the first to describe this entity. In 1808, he stated that the diverticulum comprised a remnant of the vitelline duct, the duct between the intestinal tract and the yolk sac<sup>[3]</sup>. In normal human embryology, the vitelline duct closes by the 10th week of gestation. If it persists, presentations include incidental meckel's diverticulum, fibrous cord connecting the bowel to the anterior abdominal wall, persistent omphalenteric fistula, enterocystoma, torsion, and intussusception<sup>[4]</sup>. Although the location of the diverticulum varies, they have been classically described on the antimesenteric surface of

**Table 1** Clinical characteristics, diagnostic modalities, and pathological finding of patients with inverted Meckel's diverticulum

Clinical characteristics	n (%)
Gender <sup>[4,7,9-12,14-43]</sup>	
Male	41/59 (69)
Female	18/59 (30)
Signs and symptoms <sup>[4,7,9-12,14-34,36-43]</sup>	
Bleeding	48/59 (81)
Abdominal pain	41/59 (69)
Intussusception	23/59 (39)
Anemia	47/59 (80)
Diagnostic modalities <sup>[4,7-9,12,14-34,36-43]</sup>	
Positive upper endoscopy	2/58 (3)
Positive lower endoscopy	4/59 (7)
Positive abdominal ultrasonography	12/13 (92)
Positive tagged RBC scan	0/3 (0)
Positive Meckel's scan	0/3 (0)
Positive barium enema	0/0
Positive enteroclysis	7/7 (100)
Positive upper GI series with small bowel follow through	18/21 (86)
Positive abdominal CT scan	24/24 (100)
Pathologic findings <sup>[4,7-9,12,14-34,36-43]</sup>	
Ulceration	40/59 (68)
Ectopic gastric tissue only	18/59 (31)
Ectopic pancreatic tissue only	13/59 (22)
Ectopic gastric and pancreatic tissue	4/59 (7)
No ectopic tissue	24/59 (41)

GI: Gastrointestinal; CT: Computed tomography; RBC: Red blood cells.

the ileum within 100 cm of the ileocecal valve<sup>[5]</sup>. Possible complications include hemorrhage, obstruction, diverticulitis, hernia, tumor, and inflammation, one of which an estimated 2% of those with Meckel's diverticulum will develop<sup>[1,2]</sup>. The presentation of these complications often produces a complex constellation of recurrent symptoms consistent with obstruction, chronic abdominal pain and lower gastrointestinal hemorrhage which commonly delays diagnosis and definitive surgical treatment<sup>[5]</sup>.

An inverted Meckel's diverticulum is a condition where the Meckel's diverticulum literally inverts on itself; however, the pathophysiology underlying this rare phenomenon is not clearly understood. One theory is that there is abnormal peristalsis of the bowel segment in the proximity of the Meckel's diverticulum, possibly due to the tissue present at the base of the diverticulum itself, which causes the diverticulum to invert. Because of the inversion of the Meckel's diverticulum it may be difficult for diagnostic studies which rely on access to the lumen of diverticula, such as capsule endoscopy and colonoscopy, to identify this rare lesion. This inversion of the Meckel's diverticulum can then also lead to a complete intussusception of the bowel or to a compromise in blood flow to that bowel, ulceration and then gastrointestinal hemorrhage<sup>[6]</sup>.

Because of the clinical challenge of diagnosing this rare entity, inverted Meckel's diverticulum has been reported in less than 70 cases reported in the English literature. However, as diagnostic modalities have improved the reports of this disease as an occult source of hemorrhage

has increased from 40 cases from 1978 to 2005, to 19 cases reported in the last 6 years alone (Table 1). The largest series thus far reported included 18 cases, between 1971-1995 from the Armed Forces Institute of Pathology (AFIP)<sup>[7]</sup>, and the most recent systematic review was in 2005 before the surge in reports<sup>[5]</sup>. Therefore, our report and review comprehensively reviews all these cases in the literature to further guide clinicians in the approach to the diagnosis and treatment of this readily curable disease (Table 1).

The median age of presentation of inverted Meckel's diverticulum is 27.7, slightly younger than reported by the AFIP, which was 33, with a male to female ratio of approximately 2.33:1. The most common presenting complaint was bleeding in 48 of 59 cases (80%), anemia 47 of 59 cases (78%), and abdominal pain (68%) (Table 1). With the most common presentation being lower gastrointestinal bleeding, it is not surprising that most reported cases included a thorough work up for the source of hemorrhage<sup>[5]</sup>. In most cases involving bleeding and anemia, patients underwent an upper and lower endoscopy with negative results.

The first reported radiographic description of an inverted Meckel's diverticulum was by Fetterman *et al*<sup>[8]</sup> in 1968 and there has been a great proliferation of diagnostic modalities available to clinicians<sup>[5]</sup>. A Meckel's scan, a radionucleotide scan that detects gastric mucosa, can be a useful diagnostic tool, especially in the pediatric population. It has a high sensitivity but low specificity. Only 50% of cases are believed to be associated with ectopic gastric or pancreatic mucosa, but it is seen in 75% of those presenting with symptoms<sup>[9]</sup>. Meckel scans were reported in only three cases<sup>[4,10,11]</sup> which were negative. One of these actually had both ectopic gastric and pancreatic mucosa<sup>[4]</sup>. This strongly suggests that a negative Meckel scan does not rule out the diagnosis, which was exemplified in our case.

Other radiologic diagnostic modalities include ultrasonography, which demonstrated positive or clinically influential findings in 12 of 13 cases (Table 1). These findings were often non-specific and only prompted surgical exploration in one case of a post-operative bowel obstruction caused by an intussusception from the inverted Meckel's diverticulum which was detected by ultrasound<sup>[12]</sup>. Some of the nonspecific findings include "eggplant shaped mass within the bowel"<sup>[13]</sup>, fluid filled target<sup>[14]</sup>, and distended loops of bowel with free fluid<sup>[15]</sup>. The use of a tagged nuclear red blood cell scan was reported in three cases with negative results, and barium enema has been of little use (Table 1). When a patient presents with massive hemorrhage, angiography may be useful, especially in a hemodynamically unstable patient. In a single reported case of angiography used for the diagnosis of Meckel's diverticulum, angiography revealed a vitelline artery centrally located in the ileal lumen<sup>[10]</sup>.

The three most useful tools employed for the diagnosis of inverted Meckel's diverticulum include small bowel follow-through, enteroclysis, and abdominal CT scans<sup>[4,13,16]</sup>. When the scan reveals a mass, it often is as-

sociated with a central area of fat density<sup>[5]</sup>. Small bowel follow-through was helpful in 18 of 21 cases. Findings include mass-like lesions, polypoid filling defects, and ulcerations. CT scans have been extremely helpful, especially recently with improving technology. CT scan was used in 24 of the reported cases, and all revealed useful information that ultimately led to an operation. They were especially useful when intussusceptions were found in association with the characteristic "target sign". In adults, intussusceptions with clinical symptoms are a clear indication for operation. In the pediatric population, however, even intussusceptions caused by an inverted Meckel's diverticulum can be treated non-surgically with barium enema reduction<sup>[17]</sup>. When the small bowel is highly suspected as the source of hemorrhage, enteroclysis has been suggested as the single best study in the diagnosis of inverted Meckel's diverticulum. In 7 reported cases, all seven were useful, as they revealed filling defects and polypoid lesions (Table 1).

In the literature review of the specimens, the average length of the inverted segment was approximately 3.99 cm. Ulceration has been reported in the adjacent ileal mucosa, in the Meckel's segment, and in the tip. There appears to be no direct correlation with the presence or absence of ectopic tissue. The ulceration seen in cases without gastric mucosa may be explained by either ischemia and/or trauma<sup>[5]</sup>. Fifty-eight percent of the reported cases were associated with ectopic mucosa of either gastric or pancreatic origin (Table 1).

The preferred treatment of any symptomatic Meckel's diverticulum is surgical. Whenever an inverted Meckel's diverticulum is diagnosed either pre-operatively or intra-operatively, the surgical procedure should be segmental resection with reestablishment of bowel continuity. Intussusception was noted in our case preoperatively. In our literature review, twenty three cases documented active intussusception at the time of operation (Table 1). There was one report of an endoscopic mucosal resection which resulted in iatrogenic perforation requiring emergent laparotomy<sup>[18]</sup>. It has been the general consensus that intussusceptions in the adult should be treated with resection and primary anastomosis<sup>[11]</sup>. Although most reports have described laparotomy, some minimally invasive techniques have been described in the literature. El-Dhuwaib *et al.*<sup>[14]</sup> and Karahasanoglu *et al.*<sup>[15]</sup> reported exploration and resection laparoscopically for an inverted Meckel's diverticulum. However, others have reported that manual palpation or laparoscopic inspection of the small bowel itself is not enough and may lead surgeons to miss the diagnosis<sup>[17]</sup>. In our case, we planned on an initial abdominal exploration laparoscopically, and if unsuccessful, had planned on conversion to an open laparotomy with possible intra-operative endoscopy. Fortunately, we were able to find the lesion laparoscopically and performed the resection *via* a mini-laparotomy, which has the potential to provide less morbidity than a larger laparotomy incision. Our approach provided adequate exposure to achieve appropriate margins if the

lesion had been found to have been malignant. In cases where even open laparotomy fails to localize the lesion, the successful use of intra-operative endoscopy to localize the lesion and guide treatment of inverted Meckel's diverticulum has been reported<sup>[19]</sup>.

Bleeding seen in inverted Meckel's diverticulum cannot be attributed entirely to ulceration secondary to gastric mucosa, and it may be due to trauma or inversion induced mucosal ischemia<sup>[5]</sup>. In fact, most cases did not have gastric cells present (Table 1). Trauma, due to its location within the lumen, is likely a primary source of bleeding in most cases reported in association with normal intestinal mucosa<sup>[5,13,20-22]</sup>.

There is some debate as to the treatment of incidentally discovered Meckel's diverticulum in the asymptomatic patient. Resection is generally recommended for patients younger than 40, diverticulum longer than 2 cm, divertula with narrow necks, fibrous bands, ectopic gastric tissue, and/or when the diverticulum appears thickened and inflamed. When a Meckel's diverticulum is discovered as the lead point to an intussusception, it is thought to be a primary pathologic process, and not a secondary process<sup>[5,17]</sup>. The exact cause of the inversion is not yet understood. Intussusceptions are primarily seen in children under the age of 2, and only 5% of all intussusceptions are seen in adults<sup>[1]</sup>. In children however, there is no lead point in 95% of the cases<sup>[23]</sup>.

Multiple diagnostic modalities have been described in the diagnosis of inverted Meckel's diverticulum. In instances of lower gastrointestinal hemorrhage, it is appropriate to first exclude an upper gastrointestinal source and a colonic source. Based upon a review of the literature, the studies recommended when inverted Meckel's diverticulum is suspected are CT scans and enteroclysis. However, to make this difficult diagnosis requires a high index of suspicion with an awareness of this important pathologic process and its unique presentation.

Although Meckel's diverticulum is usually an asymptomatic common congenital abnormality of the gastrointestinal tract, it can present with lower gastrointestinal hemorrhage<sup>[35]</sup>. In the case of inverted Meckel's diverticulum, the bleeding may be due to the presence of ectopic gastric mucosa, but may also be commonly due to trauma or inversion induced ischemia. In a patient presenting with lower gastrointestinal bleeding, upper and lower endoscopy can be used to rule out a source. If these modalities are negative and Meckel's diverticulum is suspected, CT scan or enteroclysis may be more helpful in the diagnosis than other modalities, and its wider use may account for the increase in reports of this rare disease in the literature. Treatment usually provides a complete cure when it entails operative resection, either *via* an open or laparoscopic approach with possible intra-operative endoscopy. Because of the non-specific presentation of inverted Meckel's diverticulum as an occult source of lower gastrointestinal hemorrhage, it is important for gastroenterologists and surgeons to understand the pathophysiology, appropriate diagnostic approach and therapeutic management of this readily curable disease.

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