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Possible Role of Meckel's Scan Fused with SPECT CT Imaging: Unraveling the Cause of Abdominal Pain and Obscure-Overt Gastrointestinal Bleeding

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Key Words

Meckel's diverticulum · Obscure-overt gastrointestinal bleeding · Chronic abdominal pain

Abstract

A 27-year-old male presented with recurrent abdominal pain and high volume hematochezia despite undergoing extensive testing and a right hemicolectomy 3 years prior for a linear bleeding ulceration in the ascending colon. Studies at the University of Michigan included esophagogastroduodenoscopy (EGD), colonoscopy and video capsule endoscopy (VCE), revealing an arteriovenous malformation (AVM) in the terminal ileum. He was hospitalized for recurrent symptoms. His presentation suggested a small bowel source of obscure-overt GI bleeding based on prior non-diagnostic colonoscopy and EGD and a bilious nasogastric lavage. Tagged red blood cell scan localized bleeding to the right lower quadrant. Colonoscopy showed fresh blood in the terminal ileum without a clear source. Angiography showed no evidence of bleeding or terminal ileal AVM. A novel Meckel's scan fused with SPECT imaging showed focal uptake in the terminal ileum. The patient underwent Meckel's diverticulectomy with sparing of adjacent bowel and has remained asymptomatic for 19 months. This case illustrates that patients with obscure-overt GI bleeding require a step-wise multi-modality diagnostic work-up. Because Meckel's scans are false-positive in 28% of adults, Meckel's scan fused with SPECT imaging may offer an approach to refine diagnostic accuracy of either scan alone, but requires further investigation. Exploratory laparotomy should be reserved as a last option and is best performed with intraoperative endoscopy.

Case Report

A 27-year-old male presented to an outside facility in 2002 for abdominal pain and massive hematochezia. Esophagogastroduodenoscopy (EGD) was non-diagnostic. Colonoscopy showed a long linear bleeding ulceration in the ascending colon leading to surgical laparotomy to perform a right hemicolectomy, but not intended to further investigate the cause of obscure bleeding. Gross pathologic examination showed an 8.1 cm linear hemorrhagic mucosal defect without evidence of acute or chronic ulceration, raising the strong possibility that the laceration was iatrogenic rather than the underlying cause of obscure bleeding. Subsequently, he developed recurrent episodes of sharp, lower abdominal pain frequently associated with nausea and vomiting followed by large volume hematochezia requiring chronic narcotics and intermittent transfusions over the next 3 years. Repeated additional tests were non-diagnostic: EGD, colonoscopy, small bowel push enteroscopy (an extended EGD), video capsule endoscopy (VCE), upper GI and small bowel studies and abdominal/pelvic CT.

He was referred to the University of Michigan in December 2005. EGD and colonoscopy were again repeated with random biopsies which were non-diagnostic. Repeat VCE revealed a non-bleeding, large 'red lesion' near the terminal ileum interpreted as an arteriovenous malformation (AVM) (<u>fig. 1</u>). His characteristic abdominal pain recurred, but was managed conservatively, and he presented with massive hematochezia 3 weeks later. He did not use aspirin, nonsteroidals, or alcohol and had no history of hepatitis or pancreatitis. Blood pressure was 115/66, heart rate 95, nasogastric lavage returned bilious fluid and Hgb was 12.3 g/dl on admission and fell to 7.8 g/dl over the following 12 h. Exam revealed no skin rashes, stigmata of chronic liver disease, telangiectasia or subcutaneous cysts.

The presentation suggested a small bowel source of obscure-overt GI bleeding based on prior nondiagnostic colonoscopy and EGD and the bilious nasogastric lavage. A tagged red blood cell scan was interpreted as showing increased tracer activity in the right lower quadrant. At colonoscopy there was fresh blood in the terminal ileum without a clear source. Angiography showed no evidence for active bleeding or terminal ileal AVM. A modified Meckel's scan, Tc-99m pertechnetate scintigraphy (fig. 2a) fused with SPECT (single photon emission computed tomography) imaging (fig. 2b, c), was performed and showed focal uptake in the terminal ileum. The patient underwent surgical diverticulectomy of a 3.2 cm Meckel's diverticulum (fig. 3a), sparing adjacent intestine, including the ileo-colonic anastomosis located 12 cm downstream. Histological sections showed ectopic gastric mucosa adjacent to duodenal mucosa (fig. 3b) and no evidence of AVM. At 19 months follow-up the patient remains asymptomatic, off narcotics without any further episodes of GI bleeding.

Discussion

Approximately 5% of patients with overt GI bleeding have a small bowel source between the ligament of Treitz and the ileocecal valve designated as obscure (negative EGD and colonoscopy) [1, 2]. The differential diagnosis is broad (see <u>table 1</u>). The most common diagnoses are AVM, ulcers and erosions [1, 3, 4], but small bowel tumors are more common in younger groups [5]. In this case, the leading diagnoses considered included AVM, intussusception and/or Meckel's diverticulum and ileo-colonic anastomosis ulceration. This patient's symptoms provided a clinical clue of possible intermittent intestinal obstruction preceding episodes of overt GI bleeding.

We have no direct evidence that the obscure GI bleeding originated from the patient's Meckel's diverticulum or evidence of hemorrhagic stigmata in the surgically resected diverticulum. Based on substantial indirect evidence, however, we argue but do not directly prove that the Meckel's diverticulum rather than the AVM or ileo-colonic anastomosis precipitated bleeding. First, GI bleeding was massive, which is atypical for AVMs. Second, colonoscopy detected no anastomotic ulcerations or inflammation. Third, surgery was a diverticulectomy that spared surrounding intestine, including the anastomosis (located 12 cm distally) and likely the AVM (not detected in the gross surgical pathology), although the latter could have been a misdiagnosis for Meckel's. Finally, the patient remains symptom free after 19 months follow-up. We speculate that

the Meckel's caused intermittent intussusception (correlating with symptoms of pain and constipation prior to bleeding) and secondary bleeding from ileal mucosal injury.

Meckel's diverticulum, first described by Fabricius Hildanus in 1598, is a 1–11 cm remnant of the embryonic omphalomesenteric duct situated 40–130 cm from the ileocecal valve and localizes to the right lower quadrant [6]. This congenital variant is present in 1–3% of the population [7] and poses a 4% lifetime risk of becoming symptomatic with GI bleeding, inflammation or obstruction [8]. Classically children present more commonly with GI bleeding adjacent to acid producing gastric mucosa and adults develop obstruction [7, 9, 10], although rarely adults older than 40 may develop bleeding. Obstruction more commonly arises by (1) entanglement of the small bowel around a fibrous cord extending from the diverticulum to the umbilicus, abdominal wall or viscera, but (2) may also occur in the free and unattached diverticulum by intussusception with the diverticulum serving as the lead point or (3) obstruction of Meckel's diverticulum by a fecalith with diverticulitis causing inflammation and adhesions [6].

The Meckel's scan involves planar, scintigraphic detection of Tc-99m pertechnetate, an anion which is intravenously infused and selectively taken up by mucous secreting cells lining gastric and ectopic gastric mucosa [6, 7, 9]. Ectopic mucosa is present in ~50%, of which 60% contain gastric mucosa [10], which increases to 90% of bleeding Meckel's diverticula [11]. Diagnostic accuracy of the Meckel's scan is >90% in the pediatric population [12] but is less accurate in adults (sensitivity 62.5%, positive predictive value 60% [13]), which is in part because complications in adults are less commonly associated with ectopic gastric mucosa [14]. This data indicates that based on the clinical suspicion, diagnosing Meckel's diverticulum should be pursued as a cause of obscure GI bleeding and may require a multi-modality diagnostic approach.

The diagnostic evaluation raised a concern that either the AVM (interpreted on VCE) or intestinal anastomosis (prior right hemicolectomy) might cause a false-positive Meckel's scan (see list of causes in <u>table 2</u>), which occurs in 28% of adult studies [13]. For this reason, the Meckel's scan fused with SPECT imaging [15, 16] was a non-invasive, diagnostic solution to refine the diagnostic accuracy of the Meckel's scan alone. The value of SPECT/CT with co-registered (fused) images is that the precise anatomic location of a focus of uptake can be ascertained. This can potentially eliminate false-positive Meckel's scans due to activity in the urinary track or vascular anomalies adjacent to bowel loops. Conversely, because most Meckel's scans are negative, the added value of CT increases the likelihood that the combined exam can offer information relevant to the patient's care and potentially reveal other causes for abdominal pain and bleeding.

Presently, SPECT/CT is not widely available and there is limited data on the utility of this imaging modality in the evaluation of obscure sources of GI bleeding. Descriptions of Meckel's scans fused with SPECT imaging are limited to two case reports [15, 16]. Compared to the standard planar imaging used to perform most Meckel's scans, SPECT imaging has additional charges, however, most insurance companies do not reimburse for this study. At our institution, there is no additional fee for the CT or fusion imaging when the CT is performed for anatomic correlation. If the CT were performed for diagnostic purposes, then additional charges would be appropriate. In the current paradigm, diagnostic CT scans are performed for the evaluation of vascular perfusion abnormalities as well as for vascular anomalies. In the future, protocols could potentially be developed to optimize both the CT portion of the study would need to be targeted to the clinical question.

This case of obscure-overt GI bleeding illustrates several additional points. VCE has a higher diagnostic yield than the combination of push enteroscopy, small bowel enteroclysis and selective angiography (68 vs. 38%) [17], but experience diagnosing Meckel's diverticulum is limited [18–22] and requires heightened clinical suspicion. Clinical suspicion should drive a step-wise work-up [23] to define endoscopic, angiographic or surgical targets. It is important to point out that double balloon small bowel enteroscopy was not available at the time the patient presented but has emerged as a valuable new tool with a similar diagnostic yield as VCE (53 vs. 65%) [4] and permits endoscopic therapy of previously unreachable small bowel lesions, possibly obviating the need for surgery. Although exploratory laparotomy is diagnostic in up to 64% of patients [24], it carries risks of severe complications (up to 12%) and mortality (up to 8%) [25] and should generally be reserved as a last option and best performed with intraoperative endoscopy, which increases the diagnostic yield to 50–100% [25], if no obvious source is found.

Charles Mayo lamented the difficulty of diagnosing Meckel's diverticulum as a persistent challenge. His laments were answered partially by the clinical implementation of the Meckel's scan in 1970 [15]. Diagnosis, however, remains a challenge, particularly in the adult patient, but may be eased further with implementation of new or modified imaging technologies to refine the diagnostic accuracy of individual studies, as used in this case.

Table 1. Small bowel causes of obscure GI bleeding

Aortoenteric fistula Angiodysplasia Crohn's disease Intussusception Meckel's diverticulum Neoplasm NSAID induced small bowel injury Polyps Small bowel varices



Case Rep Gastroenterol 2008;2:83–90 DOI: 10.1159/000119642 © 2008 S. Karger AG, Basel ISSN 1662–0631 www.karger.com/crg

Table 2. Causes of a false-positive Meckel's scan

Esophagus (Barrett's epithelium) Pe Larynx Jej Duodenum Ly Retained gastric antrum	eutz-Jeghers syndrome junal neurinoma ymphoma of small bowel
Gallbladder Vascu	ular lesions
Cystic duct He	emangioma
Common bile duct Al	bdominal aortic aneurysm
Pancreas Ai	rteriovenous malformation
Jejunum	
Duplication cyst of ileum Renal	llesions
Vermiform appendix U	reteral obstruction
Colon Ca	alyceal diverticulum
Rectum Di	ilated renal pelvis
Bladder Ec	ctopic kidney
Placenta Ve	esicoureteral reflux
Intrathoracic gastrogenic cyst	
Vascu	ular impairment
Inflammatory lesions In	itussusception
Appendicitis Vo	olvulus
Appendiceal stump Sn	nall bowel obstruction
Abdominal abscess	
Regional enteritis Other	r
Colitis Sa	acral meningomyelocele
Nonspecific small bowel ulcer In Ut	nproper identification of structures terine blood pooling

Adapted from [13] (with permission from Blackwell Publishing).

Fig. 1. Video capsule endoscopy shows an AVM in the terminal ileum.



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Fig. 2. Diagnosis of Meckel's diverticulum by imaging. **a** Meckel's scan was performed by administering 15 mCi of Tc-99m pertechnetate intravenously as a bolus. A coronal planar image shows one focus of intense tracer uptake in the left upper quadrant, corresponding to normal gastric uptake (S), and a second focus of intense uptake localizing to the right lower quadrant, suspicious for Meckel's diverticulum (M). **b**, **c** Meckel's scan images are fused with axial (**b**) and coronal (**c**) SPECT (single photon emission computed tomography) images to localize and define the abnormality responsible for the positive Meckel's scan. In the right lower quadrant near the colon (C) a focus of intense uptake localized to the terminal ileum, consistent with a Meckel's diverticulum (M).



Fig. 3. a Cross section of Meckel's diverticulum. **b** Low power microscopic examination of H&E stained specimen showing junction of ectopic gastric mucosa on the left (asterisk) with small intestinal mucosa on the right (arrow).



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