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CASE REPORT

Methylene Blue Enteric Mapping for Intraoperative Localization in Obscure Small Bowel Hemorrhage: Report of a New Technique and Literature Review

Combined Intraoperative Methylene Blue Mapping and Enterectomy

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Abstract

Background Small bowel sources of obscure gastrointestinal bleeding present both a diagnostic and therapeutic challenge. Due to the normal external appearance of the vast majority of small bowel lesions that cause obscure gastrointestinal bleeding, multiple methods of intraoperative localization have been reported. When an arteriographic abnormality is found, the use of vital dye enteric mapping is one of the most effective localization techniques.

Case Report We present a new technique combining superselective mesenteric angiography with methylene blue enteric mapping and small bowel resection performed during the same operative procedure. This technique was successfully applied in a patient with a jejunal arteriovenous malformation. Included is a review of methods of intraoperative localization with a focus on vital dye staining-guided enterectomy.

Introduction

Obscure gastrointestinal (GI) bleeding represents a diagnostic challenge to both medical and surgical teams. Exhaustive diagnostic studies can result in costly hospital expenses and risk of procedure-related complications, often without positive identification of a treatable source. Once a source of obscure bleeding is identified and determined to need surgical intervention, intraoperative localization may be equally challenging. Small bowel sources are notoriously normal in appearance during gross visual inspection, necessitating other methods of intraoperative identification. We report the case of a patient with obscure gastrointestinal bleeding secondary to a jejunal arteriovenous malformation (AVM) successfully treated via intraoperative superselective mesenteric angiography and localization by methylene blue (MB) enteric mapping allowing selective jejunal resection. A review of all cases/series of the use of vital dye mapping for obscure small bowel hemorrhage reported in the English language literature is included.

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Case

The patient was a 57-year-old female who was referred for evaluation of obscure gastrointestinal bleeding. Her past history was significant for grade two internal hemorrhoids, medication-controlled hypertension and hyperlipidemia, a prior of laparoscopic cholecystectomy, and open small bowel resection. The patient reported melena and was diagnosed with iron deficiency anemia requiring intermittent allogenic blood transfusions beginning in 2005. Her evaluation included multiple upper and lower endoscopies, multiple 99m-Technitium-labeled red blood cell scans, computed tomography, capsule endoscopy, and mesenteric angiography. In addition, due to the unclear etiology of anemia, she underwent three bone marrow biopsies, which were all negative for any

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contributing abnormality. At an outside hospital, she underwent an elective exploratory laparotomy and blind small bowel resection for suspected atriovenous malformation in 2010 after provocative angiography suggested an abnormality within the small bowel. The histopathologic evaluation of resected bowel was normal with continued melena, anemia, and monthly transfusion requirements.

Procedure

The patient was referred to our general surgery service after evidence of obscure GI bleeding continued. A full history, physical examination, review of all laboratory values, radiographic studies, endoscopic and operative reports was performed. After repeat normal upper and lower endoscopy, a literature review and consultation with the vascular surgical service were performed, and the jejunal AVM was determined to be the source of obscure GI bleeding and anemia. After counseling about the planned procedures, the patient was taken to the operating room for combined laparotomy and superselective mesenteric angiography with methylene blue injection for enteric mapping. A laparotomy revealing simple adhesions was performed while percutaneous access via the left brachial artery was obtained. Full exploration revealed no colonic or small bowel masses, lesions, or surface abnormalities. The site of previous small bowel resection was inspected and revealed dense adhesions. A self-retaining retractor was placed and small bowel positioned for inspection during intraarterial methylene blue injection.

The left brachial artery was chosen for access as it allows ease of entry into the superior mesenteric artery for endovascular procedures and allowed for concomitant laparotomy to be carried out. A micropuncture needle was used with ultrasound guidance to enter the left brachial artery, and a 4-French vascular sheath was placed for initial endovascular access. The patient received 5,000 units of intravenous heparin, the aortic arch was crossed, and the descending aorta was reached. The endovascular catheter was then positioned in the visceral aorta, and a flush arteriogram was performed. Next, the superior mesenteric artery (SMA) was selected with a hydrophilic wire and catheter, and an SMA arteriogram was performed. Advancement of the hydrophilic wire and catheter into the distal portion of the jejunal branch vessel supplying the malformation with superselective mesenteric angiography confirmed that the lesion was an AVM (Fig. 1). Palpation of the mesentery did not result in localization of the catheter secondary to postsurgical thickening and adhesions. A total of 3 mL of MB was injected through the superselective catheter. Immediate demarcation of a segment of proximal jejunum was noted (Fig. 2). Stay sutures were placed at the proximal and distal extent of color change, and 15 cm of jejunum was resected and side-to-side handsewn anastomosis performed. The specimen was inspected and opened along the antimesenteric border revealing an intense blue staining of a focal area of the mucosa consistent with active bleeding. The mesenteric defect, abdominal fascia, and skin were closed. The hydrophilic wire, mesenteric catheter, and arterial access sheath were removed without complication. The patient tolerated the procedure well and was discharged from the hospital after return of bowel function. She has had no recurrence of melena or anemia, and has been transfusion free at 12-month follow-up.

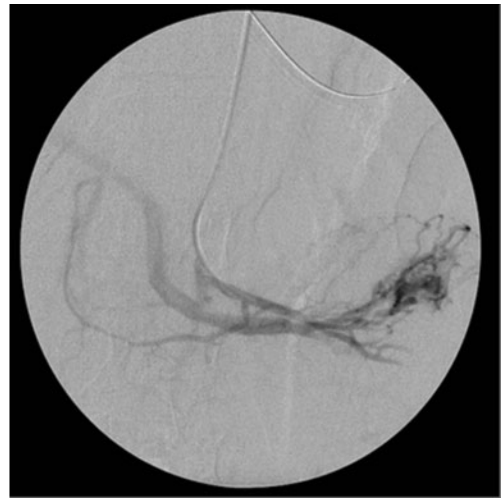

Fig. 1 Intraoperative superselective mesenteric angiogram with catheter advanced into jejunal branch confirming lesion to be consistent with arteriovenous malformation

Discussion

Obscure GI bleeding is defined as bleeding from the GI tract that persists or recurs without a clear source after endoscopic and radiologic investigation.[1] Obscure bleeding can be further categorized into obscure overt and obscure occult bleeding, determined by the presence or absence of clinically evident bleeding.[1] Despite being rare and the cause of only 1–5 % of

Fig. 2 Intraoperative appearance of intense blue staining of a 15 cm segment of proximal jejunum moments after 3 mL of methylene blue injection via a superselective catheter

all GI bleeding, small intestinal AVMs are reported to account for up to 30–40 % of cases of obscure GI bleeding in adults.[1,2] In an exhaustive review of the literature pertaining to all AVMs (n=218 reported between 1960 and 1978), Meyer and colleagues reported that 77.5 % (n=169) were found in the cecum/right colon, 10.5 % (n=23) jejunal, 5.5 % in the ileum, and 2.3 % in the duodenum.[3] This review also discussed the confusing terminology described in the literature on the subject of vascular intestinal abnormalities. Terms used to describe these lesions include: angiodysplasia, vascular dysplasia, vascular ectasia, and AVM.[3] Gastrointestinal vascular lesions can also be classified by a typing system developed by Moore and colleagues.[4] Type I lesions are found in the colon of elderly patients, type II are extracolonic lesions in younger patients, and type III lesions are hereditary hemorrhagic telangiectasias.[4] Preoperative diagnosis is most commonly made by either endoscopy or angiography.[3,5–8] Our patient's type II lesion required 99m-Technitium-labeled red blood cell scan and mesenteric angiography to diagnose (Fig. 1 and 2). Not only are colonic vascular abnormalities relatively easy to identify endoscopically but also the relatively stable fixation of the hindgut makes surgical treatment of colonic AVMs more simple than small bowel lesions.[5–7] The small bowel is not fixed or covered by the parieties compared to the colon, and typically, vascular lesions are associated with no direct readily identifiable external abnormality. This makes intraoperative techniques for localization important for successful management. Table 1 lists other reported techniques used for intraoperative localization of small bowel vascular lesions as well as limitations of each technique. Importantly, the historically described techniques of palpation, transillumination, single or multiple enterotomy(ies) for mucosal inspection, and blind resection are associated with failure in up to 50 % of cases.[9]

The use of a vital dye to map a section of small bowel containing a vascular lesion was first reported by Folger and Golemb in 1978.[10] Based on the diagnosis of a jejunal AVM made by preoperative mesenteric angiography, laparotomy was performed but upon inspection, no obvious abnormalities were noted. Using the preoperative angiogram as a guide, the superior mesenteric artery anterior (SMA) wall was exposed, and 10 mL of MB was injected directly into the artery. Resection was then guided by the finding that a segment of bowel cleared the blue dye within seconds, corresponding to the segment with rapid venous drainage via an associated AVM. Over the ensuing 34 years, a total of 25 reports of vital dye-guided small bowel resection for hemorrhage in approximately 36 total patients (one report lists the procedure without

Table 1 Intraoperative localization techniques for localization of source of bleeding

Localization technique	Limitations	References
Palpation	Vascular intestinal abnormalities are notoriously not palpable; diagnostic yield of only 30 %	33,37
Blind segmental bowel resection based on preop localization	Multiple reports of other adjuncts describe failed blind resections	37
Intraoperative assisted enteroscopy (via mouth or anus)	Not always possible to intubate and examine entirety of the small bowel; insufflates large amount of air into the lumen of bowel; cases of negative enteroscopy with subsequent positive angiographic diagnosis of AVMs with MB localization have been reported	11,33
Enteroscopy through enterotomy	See above	28,29
Enterotomy(ies) with direct mucosal inspection	Older methods of localization (blind resection, palpation, and transillumination) may miss up to 50 % of small bowel bleeding lesions	9
Intraoperative Doppler flow	Limited by lack of grossly abnormal serosal vessels; unreliable for small lesions	38 40
Intraoperative selective mesenteric venous pressure and PO_2 measurement (segment of bowel with elevated pressure and PO_2 is resected)	Useful only if enlarged veins adjacent to AVM can be identified; unreliable for small lesions; require extensive dissection of vasculature in mesentery	41
Intraoperative mesenteric angiography (selective, highly selective, and superselective)	Due to lack of posterior fixation of small bowel, difficult to precisely localize without other adjuncts	11,38,42
Intraoperative angiography with palpation of indwelling arterial catheter		38,43,44
Intraoperative mesenteric angiography with clip, platinum coil, and microcoil marking of mesentery		22,42,44,45
Intraoperative arteriography with vital dye injection	Cumbersome; risk of arteriogram (sometimes multiple) and exposure to dye; requires an angiographic abnormality	18,38

AVM arteriovenous malformation, *MB* methylene blue, *PO_2* partial pressure of oxygen clearly stating the total number treated) have been published in the English language literature.[2,11–30] Several modifications to this original technique have been developed over the years since it was first described. Angiographic modifications include the use of preoperative SMA angiography, as well as selective and superselective techniques for MB injection.[11–18] Zuckerman and colleagues described the first use of a single co-axial catheter system for superselective mesenteric angiography (SSMA) and MB injection.[17] Other dyes to include fluorescein, indigo carmine, and patent blue V dye (a dye with similar properties to MB) have been used for preoperative intraarterial injection for enteric mapping.[14,19,25,31] The volume of dye utilized has been decreased with some reports describing as little as 0.5 mL of MB for mapping.[13,14,17,19,20] Selective catheter placement with dye injection results in the staining of a segment of bowel between 10 and 40 cm in length.[13,20] The use of vital dye enteric mapping can facilitate an extremely limited resection of small bowel with Remzi and colleagues describing a 5-cm sleeve resection in a patient with only 190 cm of native small bowel proximal to an ileorectal anastomosis prior to resection.[27] Enteric mapping with vital dye has been reported in a newborn infant allowing for successful resection of a section of jejunum with bleeding ulcers.[24] Laparoscopic exploration and assisted resection has also been used in combination with preoperative MB injection.[23,29] Bowel staining with MB has been reported to be sustained up to 6 h after injection in a dog model.[32] This fact could facilitate superselective mesenteric angiography with preoperative injection of MB in patients requiring resuscitation followed by delayed transfer to the operating theater. Despite the fact that no reports describe complications of MB enteric mapping, risks of this technique do exist. Doses of 500 mg or greater of MB can cause nausea, abdominal and chest pain, altered mental status, and methemoglobinemia.[21] In addition, patients with glucose-6-phosphate dehydrogenase deficiency should not receive MB as it may cause hemolytic anemia.[21] Other than the original description of direct intraarterial MB injection into the SMA by Folger, prior reports of the use of vital dye enteric mapping have utilized preoperative angiography with transfer of the patient with a secured intraarterial catheter to the operating room. This method has the theoretical risk of catheter movement or dislodgement during transfer. To the best of our knowledge, this report is the first description of combined intraoperative SSMA with MB dye enteric mapping and small bowel resection.

Finally, one should ensure that the small bowel to be resected contains and is the source of obscure bleeding. The presence of deep mucosal staining at a single focal area confirmed that our patient's source of obscure, active bleeding was identified and treated. If no evidence of acute bleeding is evidenced by arteriography or visual inspection of the specimen, other confirmatory tests to ensure that the culprit lesion has been identified include: intraoperative enteroscopy prior to resection, completion angiography, and radiographic evaluation of the specimen ex vivo after intravascular barium injection.[19,33] Despite the most exhaustive preoperative identification of the source and method of localization, some series report that 5–37 % of patients having undergone resection of AVMs will rebleed.[3,34–36] Three possible reasons for this high rate of recurrent bleeding include: incomplete excision, possibility that the resected vascular abnormality was not the true source of bleeding, and the later development of another intestinal vascular lesion.[8]

Conclusion

Due to the normal external appearance of vascular abnormalities causing obscure GI bleeding, adjuncts for intraoperative localization of the culprit small bowel are vital. We report the first description of the combined intraoperative efforts of our vascular and general surgical services resulting in successful concurrent SSMA with MB-guided proximal jejunal resection for an AVM that failed myriad prior methods of localization and treatment. Based on our experience and the collective literature pertinent to the management of obscure GI bleeding, when surgical treatment is deemed necessary and preoperative imaging indicates a vascular abnormality, we recommend that this method be considered as it allows for optimal utilization of resources and may decrease overall procedure time and risk of complications compared to preoperative angiography and cumbersome patient transfer from angiography suite to operating theater.

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