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Coil-Localized Laparoscopic-Assisted Resection of Symptomatic Gastrointestinal Vascular Malformations in Children and Young Adults

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Background: Gastrointestinal (GI) bleeding from pediatric vascular malformation is uncommon and difficult to diagnose and manage. The preferred treatment is surgical resection; however, it can be challenging to precisely localize the lesion, particularly if it is not serosal.

Objectives: To describe a technique of intentional preoperative coil localization of symptomatic pediatric GI vascular malformations by pediatric interventional radiology to facilitate fluoroscopically assisted laparoscopic resection.

Methods: We searched the electronic privacy information center and picture archive and communication system in our center and found 3 cases. The electronic privacy information center and picture archive and communication system databases were the sources for retrieval of demographic, medical, radiological, and procedural information in all 3 cases.

Results: After many nondiagnostic investigations in all 3 patients, a GI vascular malformation as a cause of GI bleeding was diagnosed with computed tomography angiography/magnetic resonance angiography and catheter angiography. A preoperative 0.018-inch Hilal coil was placed as close as possible to the vascular malformation during super selective angiography. Laparoscopic surgery was performed within 24 hours of coil placement. In all cases, histology confirmed the resected bowel lesions to be vascular malformations.

Conclusions: Intentional endovascular coil localization has the potential to increase the precision of lesion localization and may reduce laparoscopic operative time, when guided by the coil position.

Key Words: angiography, GI bleeding, interventional radiology, laparoscopic surgery, straight coil

What Is Known

- Pediatric lower gastrointestinal bleeding from vascular malformations is rare and difficult to diagnose and treat.
- Identifying lesions during surgical resection is notoriously challenging if they are not serosal.

What Is New

- The technique described here localizes bowel vascular malformations with a coil, identifiable in the operating room using fluoroscopy for targeted laparoscopic resection.
- A potential implication of this technique increased localization confidence for nonserosal lesions being resected under laparoscopic guidance. Another implication is that coil visibility and postresection specimen radiographs with a coil in position can increase surgical confidence of complete lesion resection.

INTRODUCTION

Intestinal vascular malformations rarely present in children and are diagnostic challenges. Repeated diagnostic investigations attempt to identify pediatric gastrointestinal (GI) vascular malformations, but digital subtraction angiography is the most useful to characterize the lesion (1). Moreover, the etiology of bleeding lesions in children differs from adults. For example, angiodysplasia, the most common malformation causing GI bleeding in adults, is uncommon in children (2). In adults, transarterial embolization as a treatment for acute lower GI bleeding has high technical and moderate clinical success (3). However, embolization has limitations, such as a significant rebleed rate (reported as 19%) and bowel ischemia (5% in a series of 77 adults with lower GI bleeding) (3). Surgical resection is considered the best approach when the lesion is accurately localized; however, published pediatric reports are sparse (2,4). A prior single report has been published from our institution describing laparoscopic resection of a jejunal vascular malformation in which coil localization was used (5). This series describes in detail, with nonpublished cases, angiographic coil localization followed by laparoscopic exploration and resection, for pediatric enteric vascular lesions.

METHODS

The Institutional Review Board determined this study as "Not Human Subject Research." There were no additional institutional requirements to publish the deidentified data. A retrospective review of the electronic medical record and the radiology picture archive and communication system was performed for a series of 3 patients.

Patient demographics, clinical presentation, cutaneous stigmata of vascular malformation, associated syndromes, family history of vascular malformation, clinical management, and follow-up details were extracted and summarized.

Coil Localization Technique

All angiographic and laparoscopic procedures were performed under general anesthesia. Angiography via femoral artery access was performed by the same interventional radiologist. Glucagon was administered if bowel peristalsis degraded image clarity. First, an aortogram displayed the celiac, superior mesenteric arterial and inferior mesenteric arterial distributions. A Cobra catheter (Cook Inc., Bloomington, IN) facilitated superior mesenteric angiography. Angiographic imaging was routinely extended into the portovenous phase for venous assessment. Lesions could be identified by a hallmark "blush" (Fig. 1). Once identified, a low-flow Renegade microcatheter (Boston Scientific, Cork, Ireland) and an 0.016-inch Fathom microwire (Boston Scientific, Heredia, CR) were advanced to the feeding arcuate artery level. Next, a 0.018 inch ×

2 cm Hilal pushable straight coil (Cook Medical, Bloomington, IN) was deployed in an arcuate artery as close to the vascular malformation as possible. The straight Hilal coil design allows deployment in the most distal arcuate vessel while still maintaining configuration. The coil length of 2 cm was chosen to improve visibility under fluoroscopy. Post localization, the images were reviewed with the operating surgeon for the following: type of lesion (high versus low flow), the relationship of the coil position to the lesion (proximal, mid, distal), and the length of the vascular malformation in centimeters. After removal of the sheath, the patient was transferred immediately to the operating room, or if logistics precluded, resection was performed the next day.

Surgical Technique

All laparoscopic surgical resections were performed by the same surgeon. In general, with the patient in the supine position, a 12 mm port was placed at the umbilicus, and 5 mm ports inserted in the right upper and right lower quadrants, allowing maximum degrees of freedom to manipulate the small intestine and colon.
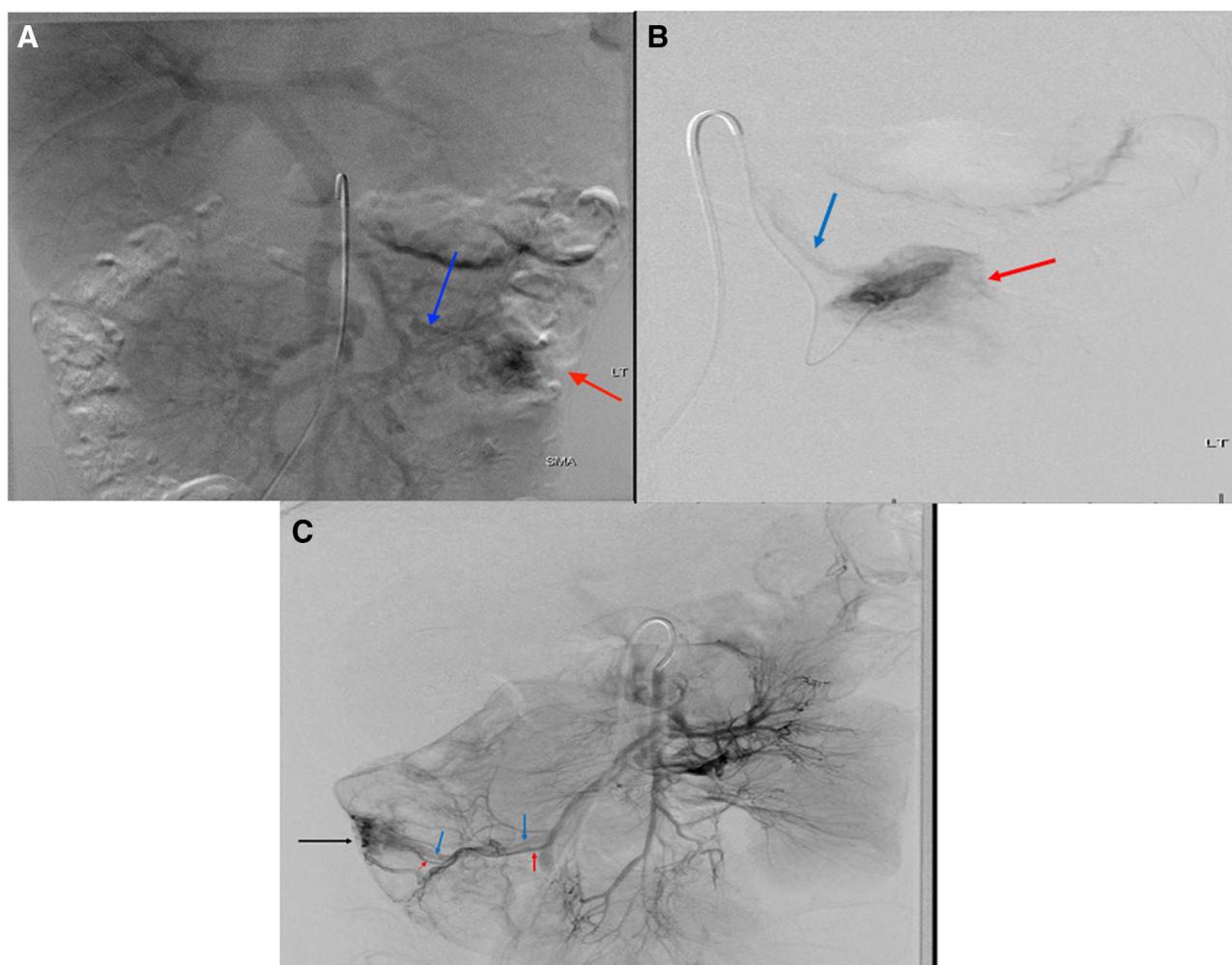
FIGURE 1. Angiography findings of patients suspected of GI vascular malformations. A 33 kg 13-year-old male with (A) portovenous phase of, SMA angiogram revealing persistent venous blush (red arrow) with dysplastic and prominent veins corresponding to the jejunal loop abnormality (blue arrow) and (B) super selective injection in the distal branch of SMA confirming venous blush with abnormal vascular dysplasia (red arrow) and dilated outflow vein (blue arrow). C) A 16 kg 5-year-old boy with right colon arterial blush in ulcer bed (black arrow) and early venous drainage into an enlarged veins (blue arrows) with corresponding arteries (red arrows). GI = gastrointestinal; SMA = superior mesenteric artery.

The small intestine was interrogated from the ligament of Trietz to the ileocecal valve, and all serosal surfaces carefully inspected. For lesions in the right colon, the cecum was mobilized along the avascular plane and white line of Toldt, and surfaces inspected. If a lesion was identified visually, the segment was manually manipulated under fluoroscopy and confirmed to be the visualized angiographic lesion by observing the localization coil at the same location (Fig. 2A, C). If the lesion was not visually apparent, fluoroscopy and coil localization were used to identify the abnormal bowel segment. Once confirmed by coil localization, the intestine segment (plus a prudent margin of ~5 cm proximally and distally) was resected with endoscopic staplers, including a wedge of mesentery containing the coil. Specimens were removed through the umbilical port site and then imaged by fluoroscopy to confirm the presence of the coil (Fig. 2B, D). All specimens were sent for pathologic evaluation. Primary reanastomosis was completed either intracorporeally or extracorporeally depending on the patient's anatomy.

RESULTS

None of our 3 cases had a family history of vascular malformations, cutaneous vascular lesions, or other stigmata of syndromic causes of vascular malformations. None of the patients underwent genetic screening for syndromic vascular malformations. The presence of GI vascular malformations was established using angiography supported by a preceding computed tomography angiography (CTA) (Cases 1 and 3) or computed tomography (CT) and magnetic resonance angiography (Case 2). Coil placement was performed electively in all 3 cases.

Case 1

A 20-year-old male with known history of congenital muscular dystrophy presented with a 7-week history of intermittent painless hematochezia and transfusion-dependent anemia. The coagulation profile and platelet number were normal. A bleeding scan, Meckel's scan, and multiple video capsule endoscopic evaluations were normal. A CTA with reconstructions demonstrated a 15 cm
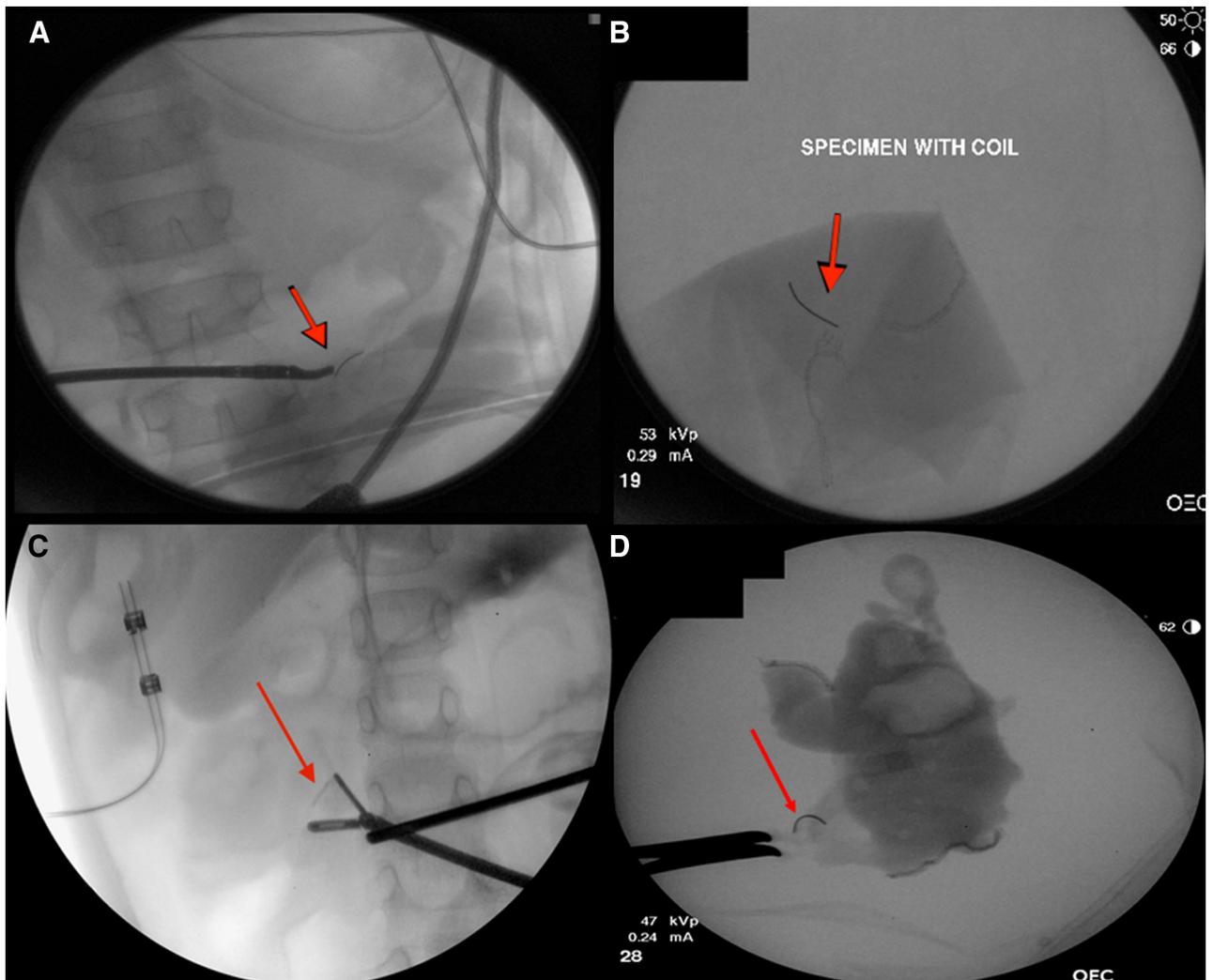
FIGURE 2. Intraoperative fluoroscopic identification of a coil placed near vascular lesion. In a 13-year-old male with a bleeding GI malformation, images from the laparoscopic surgery showing (A) intraoperative fluoroscopic assistance for determination of coil movement during bowel loop resection (red arrow) and (B) fluoroscopic image of excised bowel segment and coil (red arrow). Laparoscopic surgery in a 5-year-old male with images showing (C) intraoperative forceps engagement of the radio-opaque coil (red arrow) under fluoroscopic guidance during excision and (D) coil (red arrow) seen within a fluoroscopic image of the resected bowel. GI = gastrointestinal.

hyperenhancing loop of bowel in the left upper quadrant, likely jejunal, with dilated and tortuous submucosal veins. Digital subtraction angiography of the superior mesenteric artery and superselective injection demonstrated a subtle arterial blush in the left upper quadrant, suggestive of vascular malformation. A 0.018-inch straight pushable Hilal coil was placed just proximal to the blush (Fig. 3A) in the identified artery. Laparoscopic surgery immediately (ie, on the same day) postcoil localization confirmed that the resected specimen contained an altered venous pattern and transmural hematoma (operative time, 2½ hours) (Fig. 4A), proven to be a jejunal arteriovenous malformation (AVM) by pathologic analysis. Follow-up for 5 years revealed no recurrent hemorrhage.

Case 2

A 13-year-old male with a 1-year history of anemia of unknown cause presented to an outside institution with increasing symptoms of anemia and a hemoglobin level of 6.6 g/dL, normal coagulation panel, and normal platelet count. The anemia was managed with transfusion. Initial investigations, including esophagogastroduodenoscopy, colonoscopy, and Meckel's scan, were unrevealing. A technetium-labeled bleeding scan suggested a bleeding focus in the left upper quadrant, which on contrast-enhanced CT appeared to be in the region of a thickened jejunal loop. After transfer to our institution, sonography demonstrated a loop of markedly thickened jejunum within the left upper quadrant exhibiting increased low-flow vasculature and phleboliths, consistent with a submucosal vascular malformation. Magnetic resonance angiography confirmed this finding. The portovenous phase of the superior mesenteric angiogram demonstrated venous blush with a dysplastic vein in the jejunal loop (Fig. 1A, B). A distal arterial superior mesenteric arterial branch to this region of bowel was accessed with a microcatheter,

and a microcoil was placed to demarcate the bowel vascular malformation. Laparoscopic resection was performed the next day using the coil as a localization marker under fluoroscopy, and 10 cm of grossly abnormal jejunum was resected (operative time, 1½ hours) (Fig. 4B). Histologic examination confirmed a venous malformation of the jejunum (Fig. 4C, D). The child has remained asymptomatic on follow-up for 5 months.

Case 3

A 5-year-old boy with a history of bilateral lung transplantation due to ATP-binding cassette subfamily A number 3 deficiency (surfactant mutation), presented with a 9-month history of intermittent hematochezia, with hemoglobin level as low as 6 g/dL, a slightly elevated platelet count, and normal coagulation profile. The anemia was managed with transfusions and iron therapy. Esophagogastroduodenoscopy examination was negative. Colonoscopy demonstrated a stellate ulcer in the cecum without evidence of recent bleeding. Histologic examination of this cecal ulcer biopsy was nondiagnostic. CTA of the abdomen demonstrated abnormal vascularity in the right upper quadrant between colonic and small bowel loops without bowel wall enhancement. Superior mesenteric angiography demonstrated a persistent vascular blush in the region of the cecum/proximal right ascending colon (Fig. 3B). Selective catheterization and coiling of a distal arcuate artery adjacent to the lesion were performed prior to laparoscopic surgery immediately postangiography. The surgeon was not impressed by the physical appearance of the cecum, concluding that the lesion was likely the cecal ulcer, which was resected with the coil (operative time, 3½ hours), but the resection length was shorter than the angiographic abnormality. The laparoscopically resected ulcerated focus in the cecum with overlying thrombus was concordant with the lesion identified earlier by colonoscopy. Histologic analysis
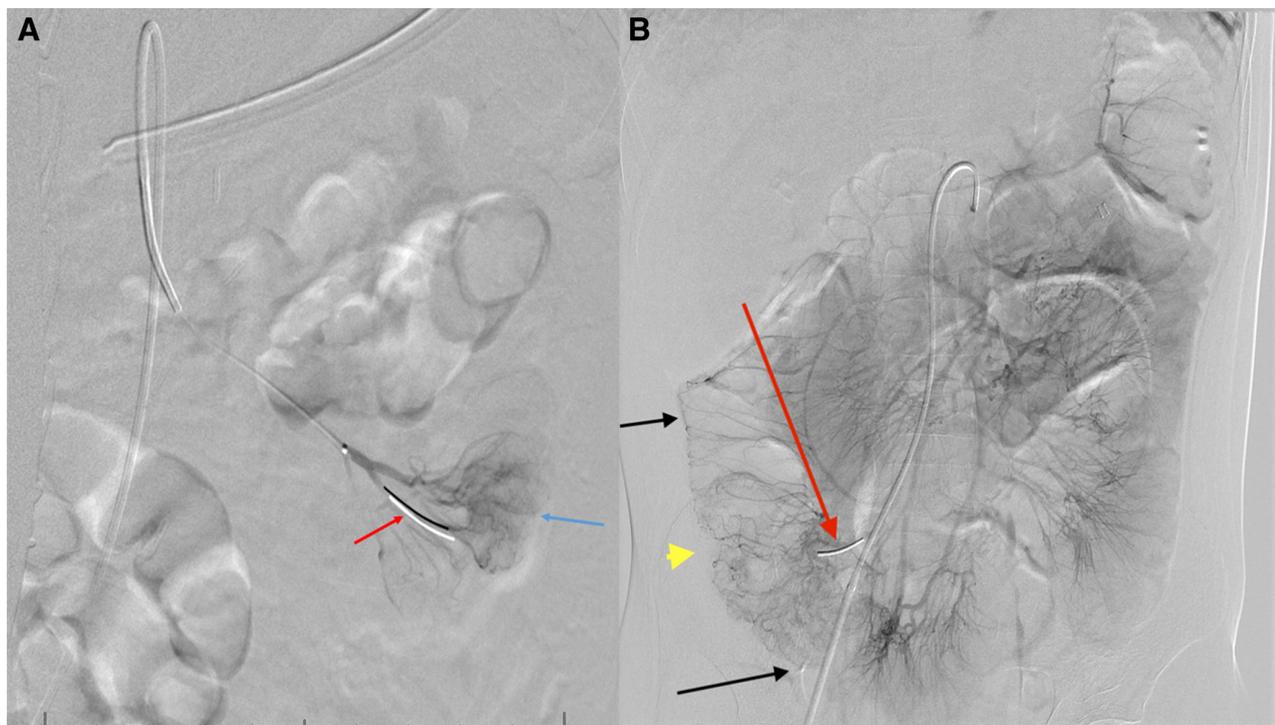
FIGURE 3. Preoperative 0.018-inch Hilal coil placement in interventional radiology suite. A) In a 50 kg 20-year-old male patient, super selective angiogram demonstrating arterial blush (blue arrow) without obvious early venous drainage and 0.018 Hilal coil (red arrow). B) In a 5-year-old male with a coil in the arcuate artery for preoperative vascular malformation localization, black arrows outlining the proximal and distal extent of the vascular malformation, yellow arrowhead marking the ulcer.

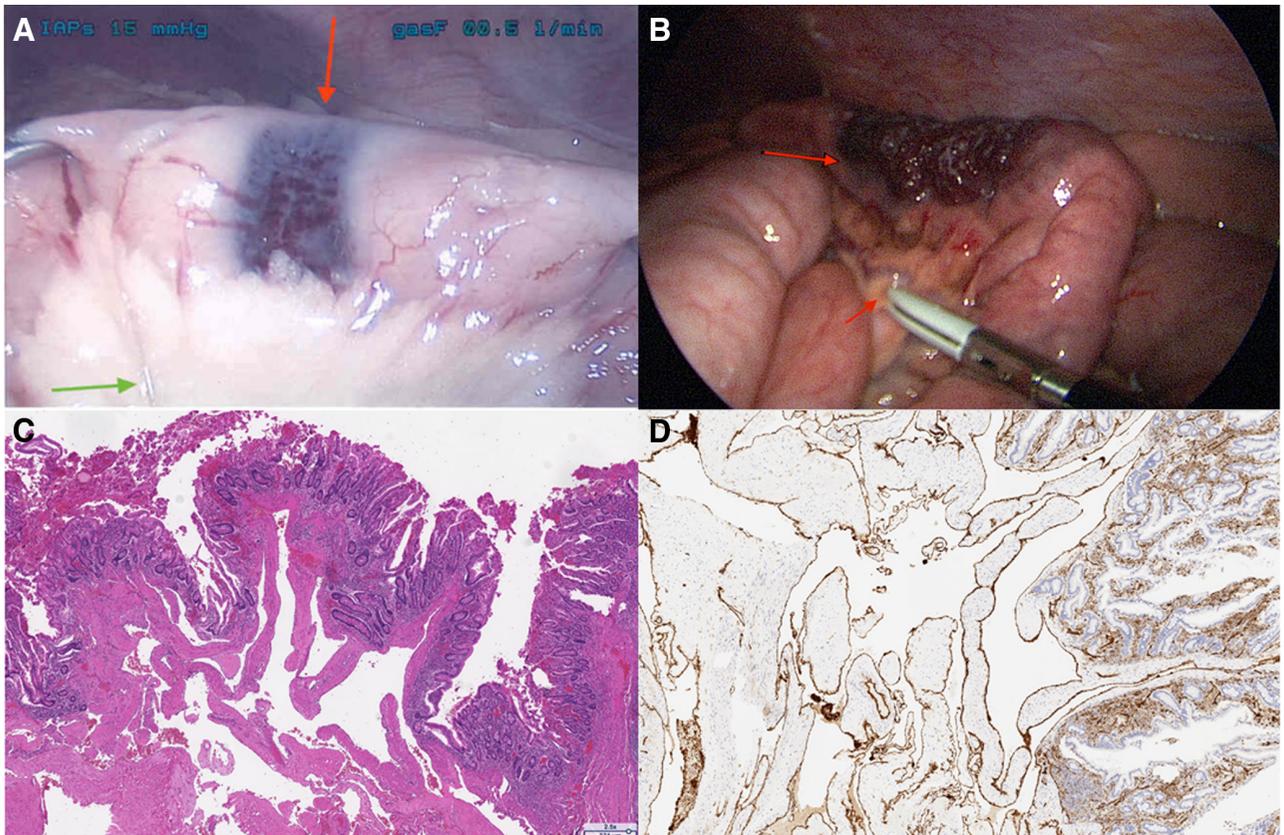**FIGURE 4.** Intraoperative and pathologic correlation. A) In a 20-year-old male patient intraoperative laparoscopic image of bowel segment with hematoma (red arrow) and coil in view (green arrow). Images from a 13-year-old patient (B) laparoscopic forceps visually grasping the site of the coil (short arrow), note the 10 cm grossly abnormal vascular lesion extending to the serosa (long arrow); (C) histology of the surgical specimen showing numerous, predominantly thin-walled vessels from submucosa to serosa with some hemorrhage and fibrin under H&E stain and 2.5× original magnification; and (D) CD31 staining was expressed strongly throughout the endothelial lining of lesion vessels, consistent with a venous malformation. H&E = hematoxylin and eosin.

confirmed the presence of an underlying AVM of the cecum. Unfortunately, intermittent hematochezia continued for another 6 months. Repeat angiography with additional coil localization demonstrated arterial tortuosity and early venous drainage, in the same region as prior angiogram, consistent with residual AVM. The patient underwent a second laparoscopic resection (operative time, 3 hours) the next day after coil placement. Histologic assessment of the excised specimen confirmed an AVM. After the second laparoscopic surgery, the GI bleeding resolved completely.

DISCUSSION

Some precedent for angiographic localization exists in adult patients. For example, intentional preoperative coil placement has been reported in 2 patients with jejunal AVMs (6,7). Coils used as a secondary localization tool at the time of laparoscopy for jejunal Dieulafoy's lesion and jejunal AVM has also been reported in adults (8,9). In these cases, the primary aim of embolization has been applied to treat acute bleeding; the embolized coils fortuitously became localizing agents when ongoing bleeding prompted operative intervention later. Preoperative CT-guided percutaneous methylene blue injection (10) and intraoperative selective angiogram with indocyanine green injection (11) have been described to assist laparoscopic resection of jejunal AVMs. In contrast, except for the prior publication from our institution, this is one of the first reports of intentional or secondary preoperative localization for GI vascular lesions in children (5).

Direct visual identification during laparoscopy or laparotomy has been reported (12–14), but the surgeon may not know in advance whether a lesion will be visible. In our series, 2 of the vascular lesions were visually identifiable during laparoscopy. In the remaining case (Case 3), the entire lesion was not clearly identifiable on laparoscopic inspection or palpation of the serosal surface; only a cecal ulcer was visible. While preoperative cross-sectional CTA imaging suggested increased vascularity between colonic and small bowel loops, without clear involvement of bowel wall, significant uncertainty remained: coil localization provided a focus for surgical exploration.

Case 3 had further intermittent bleeding postresection; a repeat angiogram identified the same distribution of high-flow vascular malformation as the prior angiogram but with evidence of the partial resection. A second coil was placed, and this area was further resected with no further occurrences of bleeding. The surgical resection included only the ulcerated area with a visible clot, but at the time, the surgeon did not identify any further abnormality on visual inspection. The need for a second operative intervention, in this case, highlights the accuracy of the initial angiographic findings compared with operative visual inspection. Placing a coil at the most superior and inferior angiographic extent may have been helpful in this case and in our practice will be considered for localization cases in the future.

In our institution, performing angiography on enteric vascular lesions prior to resection is preferable. More recently, coil localization has been added to the practice. If a lesion is clearly serosal on cross-sectional imaging, angiography and coil localization may be redundant, but the final determination of visibility is intraoperative. In our practice, adjunctive angiography and coil localization add confidence to surgical exploration. This procedure is well within the skill set of adult interventional radiologists, including most pediatric interventional radiologists. In our 3 cases, the same surgeon used fluoroscopy to localize the bowel loop, and the surgeon deemed that this significantly reduced surgical laparoscopic exploration, regardless of whether the lesion was serosal or nonserosal. In 2 of 4 procedures (Cases 2 and 3), surgical logistics precluded same-day laparoscopic resection, exposing these children to a second administration of anesthesia. One of these procedures (Case 3) was a repeat resection after the angiography, and localization resulted in a cure for this patient. Ideally, the goal is to perform localization and resection on the same day.

Coil localization has potential downsides. Some interventional procedural time is added by coil placement. In our experience, the placement of a coil in the distribution of the vascular malformation added a relatively small amount of procedural time to the angiography (mean 22 minutes, range 11–38 minutes). Additionally, radiation dose is a consideration especially in small patients. The radiation dose ranged from 40.7 to 72 mGy, relatively low doses for abdominal intervention in children. The intraoperative fluoroscopic dose ranged from 0.65 to 1.78 mGy, very low doses. Transportation to the operating room under anesthesia has inherent risks such as endotracheal tube dislodgment; hence, a hybrid interventional radiology-operating suite would obviate this problem.

Overall, we describe a combination of intentional endovascular coil localization for laparoscopic excision of enteric vascular lesions with the assistance of intraoperative fluoroscopy. This combined technique may offer more confident localization of lesions, avoids laparotomy incisions, and may decrease intraoperative time. This procedure may be considered especially in situations where lesions may not be clearly serosal or easily localized on cross-sectional imaging.

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