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Case Report

Hematochezia in a 1.5-Year-Old Child Diagnosed as a Colonic Arteriovenous Malformation

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ABSTRACT

Vascular anomalies are an uncommon cause of gastrointestinal bleeding in childhood. These malformations may present with diverse symptoms, while gastrointestinal bleeding is the most common. Regarding diagnosis, these lesions may be detected by endoscopy, but only the mucosal component of them is assessable by this modality. Definitive diagnosis is best achieved by selective mesenteric angiography. We report a rare case of a 1.5-year-old child who presented with the complaint of hematochezia, which, after examination and evaluation, was found to be due to an intestinal arteriovenous malformation. Therapy of these malformations includes different modalities such as coagulation, embolization and surgical resections. Reviewing the literature, this is the first report in pediatric surgery literature of the laparoscopic approach for symptomatic high flow arteriovenous malformation, especially at this incredibly young age.

Introduction

Vascular anomalies are rare causes of gastrointestinal bleeding in childhood. When present bleeding is the most common symptom of both hemangiomas and arteriovenous malformation (AVM). We report a case of a child that presented for the first time to our pediatric clinic in "Soroka University Medical Center" (SUMC), at the age of 1.5 years with hematochezia, which was described as painless rectal bleeding. We describe the evaluation she went through her diagnosis and treatment.

Case Report

A 1.5-year-old girl was admitted to our pediatric department in SUMC, with the complaint of rectal bleeding. The parents described that in the past few months, prior to their admission, their daughter had about 5 bowel movements a day, most of them with fresh blood. There was no history of fever, vomiting, pain at defecation or passage of mucus along with her stools. There was no history suggestive of intestinal obstruction. The child was healthy until now, her growth was normal (50 percentiles for height and weight) for age and she had a good appetite. There was no personal or family history of gastrointestinal or bleeding disorders. Physical examination at her presentation was unremarkable and she was stable hemodynamically. Her abdomen was soft with no tenderness, her skin wasn't pale and no naevi, cutaneous or mucosal telangiectasia were found. The rectal examination was negative for fissures or hemorrhoids. Her blood tests were in the normal values with hemoglobin of 12.3 gr/dL, normal INR, electrolytes and liver function tests. Her albumin was 4.8 gr/dL and CRP was 1 mg/L. Her stool was soft with fresh blood; under microscopic examination, there were many RBC with no leukocytes. Stool culture and PCR for viruses were negative.

The child went through a colonoscopy which macroscopically didn't show any pathology including polyps, but biopsies were hard to take due to haemorrhage and fragmentation of the tissue. Biopsies taken from

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different segments of the colon and ileum didn't show any pathology. The child underwent CT-Enterography of the abdomen that demonstrated diffuse thickening of the colon from the rectum till the ileocecal valve. The main differential diagnosis was between an inflammatory process and a vascular one. Due to negative inflammatory markers with repeated episodes of bleeding, it seemed more likely to be a kind of a vascular malformation. The child was lost from follow-up for a year.

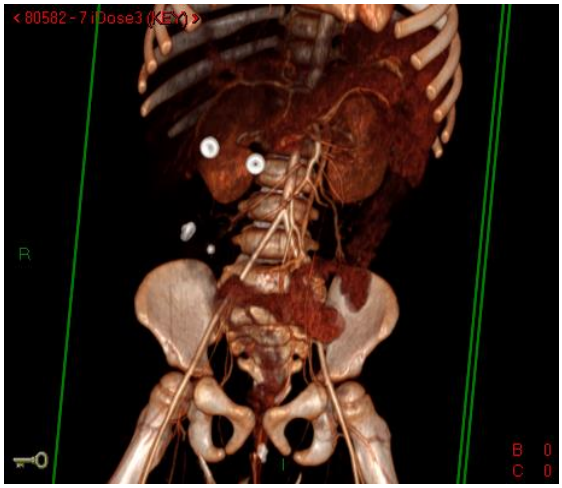

Figure 1: Coronal 3-D reconstructive image demonstrating enhancement, depicted in red, of the transverse colon through the rectum. Additionally, there is a demonstration of the enlarged inferior mesenteric artery and early filling of the inferior mesenteric vein which is noted to be enlarged.

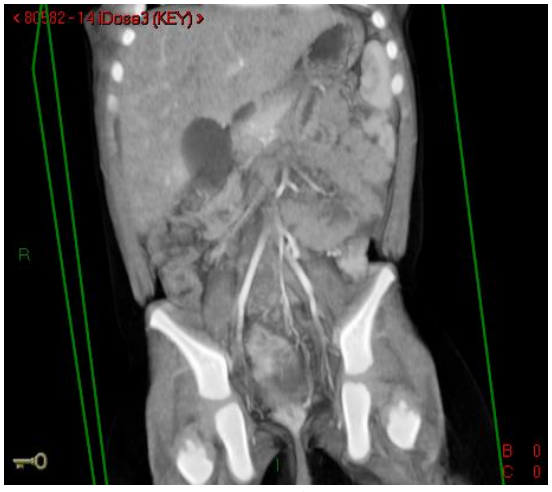

Figure 2: Mid-coronal image of the abdomen shows the enlarged feeding artery of the inferior mesenteric artery.

At the age of 2.5 years, she came back to our department with the same complaint of painless rectal bleeding. Physical examination at her acceptance revealed a pale child with no rash, petechiae or hematomas on her skin or mucus membranes. Her abdomen was soft and not tender with no organomegaly. Auscultation to her heart sounds revealed a new systolic murmur 2/6. The remainder of the physical examination was unremarkable. Her pulse was 130 bpm, BP 107/76 and the body temperature was normal. Her blood count revealed anemia with a hemoglobin level of 6.8 gr/dL with low indexes and a broad RDW. The leukocyte and platelet count were normal and chemistry including kidney, liver and pancreatic functions, were all in the normal range. Due to symptomatic anemia, the child was treated with packed cell transfusions and her hemoglobin level increased to 12.3 gr/dL. To find out the cause of her hematochezia, the child went through a CT-Angiography that showed a high flow AVM, between the inferior mesenteric artery and vein (IMA and IMV) (Figures 1-3).

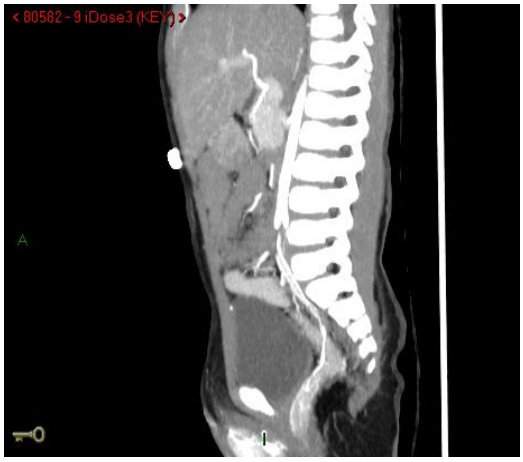

Figure 3: Sagittal midline images show the enlarged feeding inferior mesenteric artery.

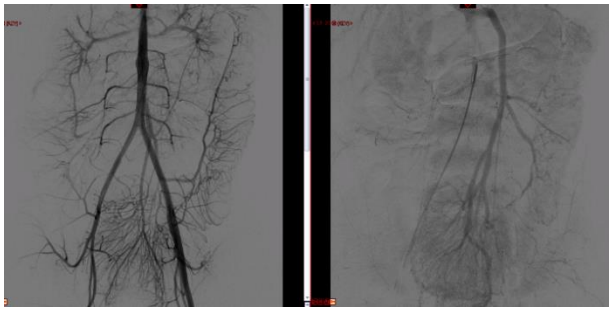

Figure 4: Angiographic images from arterial and early delays show the enlarged feeding vessel inferior mesenteric artery as well as the enlarged inferior mesenteric vein with early drainage prior to filling of the portal vein compatible with arteriovenous shunting.

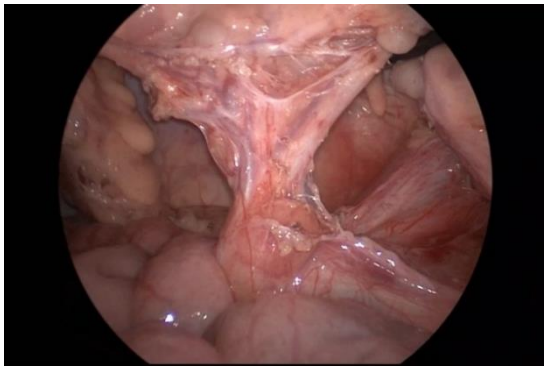

Figure 5: An AVM shunt located at the level of IMA and IMV.

Three months later, the child was referred to another center with experienced invasive radiology service for the attempt of diagnostic, therapeutic catheterization and option of selective ablation of the AVM. The angiography demonstrated a diffuse AVM, as demonstrated in (Figure 4). Selective ablation was technically unreachable and major concern was post-procedural bowel ischaemia and necrosis. The child continued to pass fresh blood in her stool and her hemoglobin level

decreased. Due to ongoing intermittent and symptomatic episodes of hematochezia, we repeated multidisciplinary consultations in the pediatric division of SUMC and concluded that the child should go through surgery-division of the AVM with an unavoidable option of left hemicolectomy. Open conventional abdominal surgery was discussed and despite the leak of the reports in the pediatric surgical literature, laparoscopic approach was our first choice.

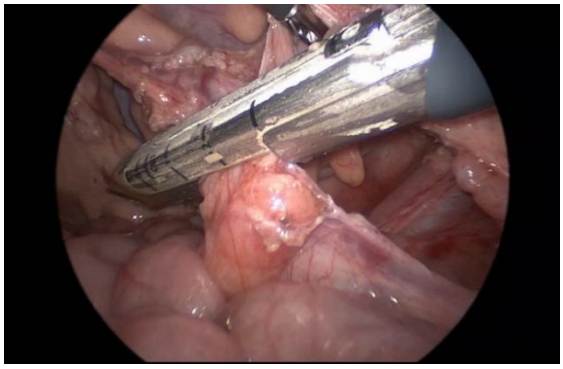

Figure 6: Stapler disconnection of persisting AVM.

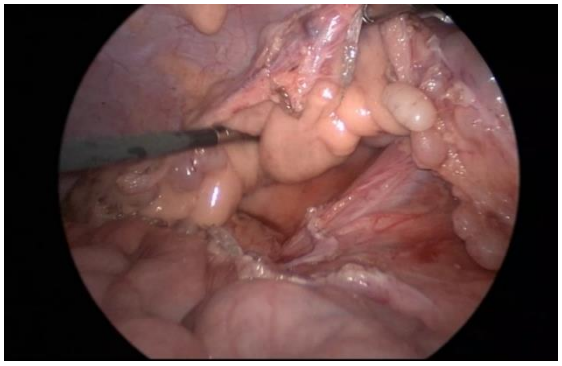

Figure 7: After clear demarcation left hemicolectomy was completed.

At the age of 2 years and 10 months and after detailed explanations-consent for surgery was obtained from the parents. The child underwent diagnostic laparoscopy. Meticulous dissection in bowel mesenterium revealed anatomical blood supply and a direct AVM shunt was located and clearly demonstrated at the level of IMA and IMV (Figure 5). Laparoscopic dissection and mobilization of the left-sigmoid and upper rectum were performed with LigaSure dissector and after stapler disconnection of persisting AVM (Figure 6) with clear demarcation left hemicolectomy was completed (Figure 7). Bowel (sample) evacuation was done through small suprapubic incision and end to end trans-rectal colorectal anastomosis was performed with EEA stapler device 6 cm above the dentate line. The operation underwent with no complications, remarkable fast recovery with minimal pain medications and excellent cosmetic outcomes. Bowel movements appeared on 3rd PO day and she reached full feeding at 5th PO day. Rectal bleeding symptoms resolved and her hemoglobin level at the day of discharge was 12.6 gr/dL.

Discussion

Many classifications of intestinal vascular malformations exist, confusion persists as different labels are given to similar lesions. The Mulliken classification for superficial vascular anomalies in children is logical and useful [1-3]. It is based on biological properties that dictate clinical evaluation. This classification distinguishes between hemangiomas, which are proliferative tumors that can regress spontaneously and vascular malformations (VM). This classification system has continuously been revised most recently in 2014 [4]. In contrast to vascular tumors, e.g. childhood hemangiomas, intestinal VM are characterized biologically by the absence of growth or by spontaneous regression. These are congenital dysplastic vessels resulting from errors in vascular morphogenesis [5]. VM can be further subdivided into low- and high- flow malformations, which would reflect their capillary- venous and arterial origin, respectively [6].

Vascular anomalies afford complex diagnostic and therapeutic challenges when gastrointestinal (GI) manifestations are present. Manifestations include GI bleeding, obstruction, diarrhea, ascites, pain, emesis, ileo-ileal intussusception, protein-losing enteropathy, and hypersplenism. Furthermore, patients can have manifestations of portal hypertension, associated musculoskeletal or cutaneous lesions, congestive heart failure, partial anomalous pulmonary venous return, pulmonary edema, and pleural or pericardial effusion. Nevertheless, bleeding is the most common symptom of both hemangiomas and AV malformations [7].

Although VM may be detected by endoscopy, only the visible mucosal component of the VM is assessable by this modality. Definitive diagnosis and determination of the site and extension of an intestinal VM are still best achieved by selective mesenteric angiography. Involved vessels and flow pattern, as imaged by angiography, are used to classify VM into low- and high-flow VM [8, 9]. Regarding therapy, several methods, including coagulation and embolization, have been used to treat vascular lesions; nevertheless, surgical resection offers a suitable therapeutic modality. While coagulation and embolization are suitable options for treatment regarding VM of solid organs, using these modalities in hollow viscus puts them at risk for ischaemia and necrosis. While right hemicolectomy is the most common surgical procedure performed in adults with vascular anomalies of the colon, in children, the left hemi-colon and rectum are the more frequently affected segments, rendering surgical therapy a challenge [10].

Conclusion

The presented case demonstrated a unique cause of lower GI bleeding, hematochezia in a child. In the pediatric population, there is limited experience regarding angiodysplasia/ AVM of the GI tract. To our knowledge, this is the first report in pediatric surgery literature of laparoscopic approach for complex/symptomatic high flow AVM (IMA-IMV shunt) and this technique may be considered in the future as a primary approach in selected cases.

Conflicts of Interest

None.

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