Clinical Case Study

OPEN

DIFFUSED VASCULAR MALFORMATION OF THE ENTIRE COLON: UNUSUAL ETIOLOGY OF GASTROINTESTINAL BLEEDING IN PEDIATRICS

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The causes of gastrointestinal (GI) bleeding in pediatrics are diverse. Unlike Meckel's diverticulum, anal fissure, polyps, inflammatory bowel disease, and infectious colitis, vascular malformation is not typically considered an etiology of GI bleeding in pediatrics, which can easily cause a delay in diagnosis or be misdiagnosed as reported (Abdoon, 2010; Al-Mehaidib, Alnassar, & Alshamrani, 2009). Vascular malformation, as an important cause of lower GI bleeding, is usually diagnosed in the elderly (Sami, Al-Araji, & Ragunath, 2014). It can affect any segment of the tract, usually the cecum and right colon in adults (Nishimura et al., 2015). Previous studies on children have shown that segmental lesions in the GI tract were found (Abdoon, 2010; Al-Mehaidib et al., 2009; Chuang et al., 2011; de la Torre Mondragón, Gómez, Mora Tiscarreño, & Ramírez Mayans, 1995; Uhlig, Stephan, Deutscher, Kiess, & Richter, 2004), but involvement of the entire colon is rare. In this case, we report an Asian boy presenting with GI bleeding diagnosed as diffused vascular malformation of the entire colon.

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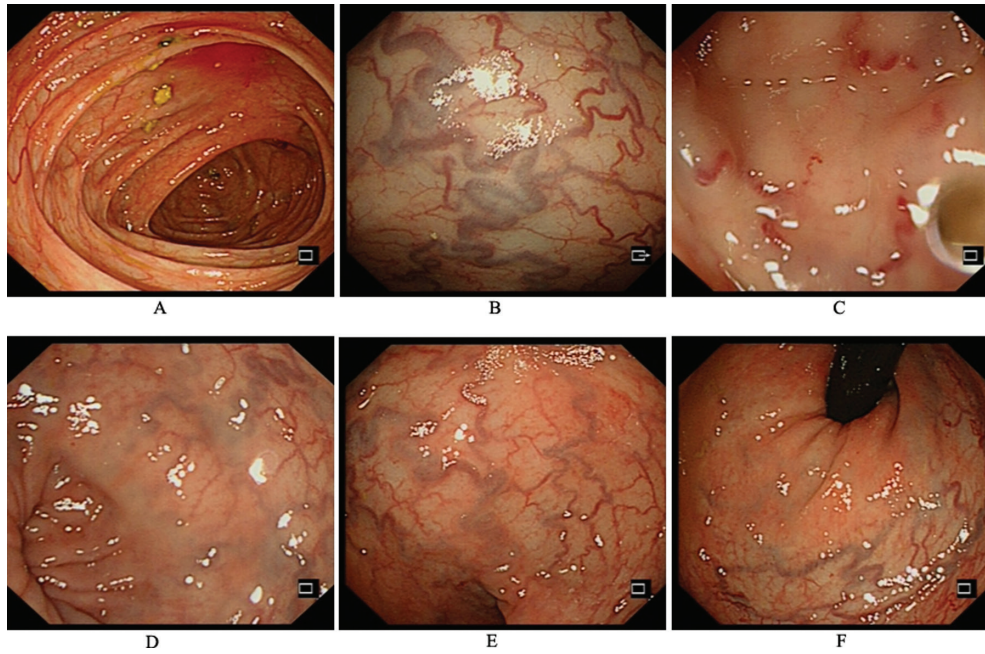

FIGURE 1. Colonoscopy of vascular malformation in different sites of colon. (A) The ascending colon. (B) The transverse colon. (C) The descending colon. (D) The sigmoid colon. (E) The rectum. (F) The anal tube.

Case

A 13-year-old Asian boy was admitted into the hospital due to discontinuous hematochezia for 2 years and abdominal pain for over 1 month. The boy had no history of disease, surgery, medication, or family history. There was not much blood in his stool and bleeding would stop spontaneously. Either bright red or dark red bloody stools were seen in the course of his disease.

At physical examination, he was generally in good condition, but had had a mild anemic appearance and complained of mild tenderness across the entire abdomen. The palpation of his abdomen was soft with no venous exposure. No hemorrhoids were found and no other abnormalities were observed. Blood routine examination showed the hemoglobin level decreased slightly as 92 g/L, and other parameters, such as white blood cell count, neutrophils, red blood cell count, and platelet count, were all within the normal range. Different tests were performed to determine the presence of the following pathogens: test for urine routine, coagulation routine, liver function, kidney function, and stool parasite were normal. Antibodies to hepatitis B/C viruses, *Treponema pallidum*, and HIV were all negative. The levels of ceruloplasmin and alpha fetoprotein were both normal.

The ultrasonography of the intestines, liver, spleen, kidneys, and heart were without any abnormalities. Furthermore, under gastroscopy, no thickened vessels were seen in the esophagus and gastric fundus. Colonoscopy showed normal mucosal but with blood vessels dilated, tortuous, and thickened throughout the entire colon (Figure 1). The terminal ileum and ileocecal valve were normal. Capsule endoscopy was also performed showing no abnormalities of the small intestine.

The pathological results suggested that there was a little infiltration of lymphocytes and plasma cells in the intestinal tract with eosinophils 2–3/hpf. Therefore, vascular malformation was considered an etiology of the GI bleeding. To further clarify the bleeding site, digital subtraction angiography was recommended but the boy did not obtain his parents' permission. The boy was totally freed from hematochezia and abdominal pain after using octreotide for 5 days and was discharged.

Discussion

The frequency of vascular malformation of the colon as a cause of lower GI bleeding is 0.6% among adults in a recent study (Tsai et al., 2018) and is unknown for children. In adults, advanced age, especially over 60 years old, heart disease, use of anticoagulant drugs, etc. have been considered risk factors of vascular malformation (Nishimura et al., 2016), whereas scarce data have been identified in children. The mean age of clinical onset was 2.3 years and average delayed diagnosis was 2.9 years (de la Torre Mondragón et al., 1995).

Vascular malformation can be incidentally found with various clinical manifestations, of which GI bleeding is the most common problem. Patients may present with chronic and recurrent bleeding, as was in our case. In previous studies, lesions were segmental, and any segment of the GI tract can be affected

(Chuang et al., 2011; de la Torre Mondragón et al., 1995; Uhlig et al., 2004). However, the boy in our study presented with diffused lesions of the entire colon, which is rarely reported.

Endoscopy and angiography are widely used in diagnosis of vascular malformation, of which endoscopy is the main tool (Sami et al., 2014). At the time of endoscopy, prominent lesions might be visualized and treated with endoscopic therapy. However, there are limitations; for example, lesions or bleeding sites can be missed during endoscopy due to a variety of reasons, such as poor visibility, size, and location of lesions (Sidhu, Sanders, Morris, & McAlindon, 2007). Angiography, as a supplemental tool, could be used to locate lesions or bleeding sites. In our case, the boy underwent endoscopy but did not undergo angiography without his parents' permission. We were unable to identify the bleeding sites, but the boy recovered well after taking octreotide. Due to the lack of evidence on outcome and treatment of vascular malformation, it is suggested that the disease management should be individualized depending on the lesion site, severity of bleeding, and general impairment.

Conclusion

This case highlights that vascular malformation should be kept in mind when dealing with GI bleeding, even in children; especially when patients have recurrent bleeding. Endoscopy is an essential tool for making this diagnosis.

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