Multiple Polypoid Angiodysplasia with Obscure Overt Bleeding: A Case report

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Abstract

Angiodysplasia (AD) is increasingly recognized as a major cause of gastrointestinal bleeding. Morphologically flat lesions are common types of AD, leaving the polypoid types rare to find. We report a case of multiple polypoid AD in the small bowel causing severe anemia and requiring surgical treatment.

A 60-year-old male visited the hospital with dyspnea and hematochezia. He had a history of myocardial infarction with taking both aspirin and clopidogrel. Capsule endoscopy, enteroscopy, computed tomography and angiography revealed multifocal vascular lesions with polypoid shape in the jejunum. Surgical resection was performed, since endoscopic treatment was considered impossible with the number and the location of lesions. The risk of recurrent bleeding related to antiplatelet agents also supported performing surgery. AD was histologically diagnosed from the surgical specimen. The patient resumed taking both aspirin and clopidogrel after surgery. He was fully recovered and has been doing well during the several months follow up.

Introduction

Angiodysplasia (AD) is the most common vascular lesion in the gastrointestinal (GI) tract. Although the cecum and the ascending colon are the most common sites of AD, 15% of AD is speculated to reside in the small bowel. It has been demonstrated that bleeding in the colon is mild and spontaneously resolving up to 90% of patients. However, lesions in the small bowel can cause occult yet severe GI bleeding which is fatal to patients.

Vascular lesions in the GI tract are one of the important causes of GI bleeding. They are generally categorized as neoplastic lesions and non-neoplastic malformations. Due to their overlapping characteristics, the classification of vascular lesions requires various modalities including endoscopic, radiologic and histologic evaluations. Neoplastic lesions include hemangioma whose endoscopic findings show polypoid, compressible and bluish feature. AD and arteriovenous malformation (AVM) belong to non-neoplastic malformation.

We here report a case of multiple polypoid AD in the small bowel which caused massive bleeding and eventually required surgical resection.
Case report

A 60-year-old male was admitted to our hospital with a two-day history of exertional dyspnea. The patient’s vital signs were stable. There were no accompanying respiratory symptoms and abnormal breath sound on auscultation. His chest x-ray was normal but laboratory findings showed severe anemia. The initial hemoglobin level was 6.3 g/dL and the hematocrit level was 20.0% with the mean corpuscle volume of 98.0 fL. For ten years since he had percutaneous coronary intervention for acute myocardial infarction, he had been taking aspirin and clopidogrel. He stopped taking both pills to avoid potential obscure bleeding. However, there was no evidence related to active bleeding or bleeding tendency on history and physical examinations. Blood test results were normal except for anemia and echocardiography showed no other abnormal findings related to the patient’s cardiovascular condition.

An Esophagogastroduodenoscopy found several erosions and small healing ulcers on the antrum. The colonoscopy result was normal. These findings were not sufficient to explain severe anemia. For the small bowel evaluation, capsule endoscopy was performed. It showed active bleeding from the proximal jejunum, but the exact mucosal lesions were not determined (Fig 1). Enteroscopy for diagnostic and therapeutic purposes was planned but could not be performed immediately because of the higher risk of bleeding related to antiplatelet agent use during the procedure. The patient stopped taking both aspirin and clopidogrel to prevent further bleeding. CT angiography revealed multifocal and small enhancing nodular lesions in the jejunum on portal venous phase (Fig 2). Mesenteric angiography showed dysplastic vessels accompanying early filling drainage at branches of the cecal and proximal jejunal artery. Multiple slow-filling nodular stains in the small bowel were also detected. Based on these CT and angiographic findings, the small bowel lesions were presumably diagnosed as unusual presentations of AD or hemangioma. During workup, the patient had packed red blood cells (RBC) transfused. The level of hemoglobin went up to 9.3 g/dL and remained stable. Dyspnea was relieved with stable vital sign. He was discharged with the instruction to take aspirin only.

Three weeks later, the patient revisited our emergency room with massive hematochezia. The hemoglobin level was 5.7 g/dL and the hematocrit level was 18.8% with the mean corpuscle volume of 92.6 fL. Accordingly, the patient stopped taking aspirin and received packed RBCs transfusion again. CT angiography was performed, and it showed multiple (>20) and small enhancing nodular lesions in the small bowel without active arterial
bleeding. Because there was no evidence of active arterial bleeding, embolization was considered not effective. Single balloon enteroscopy was performed to evaluate the recurrent bleeding in the small bowel. It revealed multiple variable-sized bluish vascular lesions with polypoid shape between the proximal and distal jejunum (Fig 3). This finding was unusual for AD so other vascular anomaly such as AVM or hemangioma was suspected first. Endoscopic treatment could be not implemented based on the number and the location of lesions.

The patient continued to bleed and received average of two units of packed RBCs per day. We decided to perform surgery for the small bowel vascular lesions. During the surgery, enteroscopy was performed and found bleeding focus. However, there was no active bleeding. The small bowel was resected from 10 cm below the Treitz ligament, resulting in 120 cm in length of the bowel resected. The specimen includes 16 polypoid submucosal hematomas with the largest measuring 1.5 x 1.3 x 0.8 cm (Fig 4). During a pathologic diagnosis, hemangioma was initially suspected grossly. However, H&E stain showed that the polypoid lesions consisted ecstatic vasculature with organizing thrombi in submucosa and elastic stains confirmed that the dilated and tortuous vasculature were composed of arteries and veins with direct communication (Fig 5). Based on the information above, multiple polypoid AD was diagnosed. He was fully recovered from surgery, and resumed taking both aspirin and clopidogrel. He has been doing well without further bleeding during the several months follow up.

Discussion

AD is usually diagnosed based on its typical endoscopic findings, bright red, 5 to 10 mm in diameter and fern-like appearance with flare-radiating peripheral blood vessels. Because AD usually has a flat or slightly raised lesion, polypoid lesion is morphologically rare, and therefore not often discussed. Out of the 17 previous case studies on polypoid AD, the most dealt with a single lesion with exception of only one case with multiple polypoid lesions. It is considered a congenital error that occurs during the development of the embryonic vasculature. Moreover, a mechanical factor may play a role in its occurrence in some cases, with vigorous peristalsis or increased intraluminal pressure causing or increasing the shunting of the blood into the submucosal arteriovenous system.

CT angiography is a potentially crucial noninvasive test because it could detect extravasation from AD.
Selective mesenteric angiography is a useful diagnostic technique for AD as well, especially when patients suffer from massive bleeding which hinders an endoscopic approach. There are three significant angiographic signs in the mesenteric angiography of AD. The earliest and the most common sign is an appearance of vein densely opacified, dilated, slowly-drained within the intestinal wall. As the lesion progresses, a vascular tuft can be found in the arterial phase. The late sign is an early-filling of vein in the arterial phase, which signals an intensified arteriovenous communication throughout the angiodysplastic lesion. Biopsy is not usually recommended due to the risk of bleeding during routine endoscopy, however, microscopic examination of AD have revealed collections of dilated, tortuous arteries and veins in the submucosa.

Although the terms AD and AVM are used synonymously due to their similar endoscopic and angiographic findings, AD is distinguished histologically and clinically from AVM. AVM is an interconnection of aberrant arteries and veins with thick hypertrophic walls. These lesions are usually congenital and tend to be found in younger patients. In contrast, AD has thin-walled submucosal vessels that are dilated, often lined by endothelium only. AD represents a degenerative process from intermittent or partial obstruction of submucosal veins, leading to capillary dilation, which results in an arteriovenous communication. Therefore, AD usually occurs in elderly patients and is predominately developed in the right colon.

Hemangioma in the small bowel is also very rare and can be divided into capillary, cavernous and mixed form. Endoscopy revealed soft compressible submucosal lesions with a polypoid shape in bluish or deep-red color. Histologically, this lesion is distinct from AD or AVM. In the simplified classification of Mulliken and Glowacki, the important difference between hemangioma and vascular malformation lies in endothelial proliferation.

In this particular case, we detected multiple vascular lesions with polypoid shape and bluish color by enteroscopy and multiple nodular stains using the CT. Based on these findings, small bowel hemangioma was first suggested. However, there was no vascular proliferation lined by endothelial cells suggesting hemangioma in pathologic evaluation in our case.

The decision whether to treat AD depends on the clinical situations. AD detected during endoscopy screening does not require a treatment, but, patients with obscure occult or overt bleeding are recommended to be treated for AD. Endoscopic treatments such as argon plasma coagulation, electrocoagulation, or photocoagulation using Nd:YAG are the available modalities for AD treatment. For those who cannot undergo endoscopic
treatment, transcatheter angiography and embolization are encouraged as alternative treatments. With the improvement of diagnostic and therapeutic modalities, the need to perform surgery decreased. However, our patient received surgery, because the given number and the location of lesions, ten polypoid lesions between the proximal and distal jejunum, obstructed endoscopic treatment. The repeatedly required transfusion also influenced our decision to perform surgery. His preexisting and ongoing obligations, such as taking aspirin and clopidogrel for coronary disease, also supported such decision. The co-morbidities and clinical situations of patients should be weighted in treating AD.

We report an extremely rare case of multiple polypoid AD which caused overt obscure GI bleeding. Endoscopic, radiologic and histological evaluations each has an important role in the diagnosis and treatment of AD. Among the treatment modalities, surgery can be considered when endoscopic or angiographic treatment is not suitable or when patients have poor prognosis factors.

Conflicts of Interest

The authors have no financial conflicts of interest.

References


Figure legends

Fig 1. Capsule endoscopy showed active bleeding from the proximal jejunum, but could not detect exact mucosal lesions.

Fig 2. CT angiography showed multifocal small enhancing nodular lesions in the jejunum.

Fig 3. Single balloon enteroscopy showed multiple variable-sized bluish vascular lesions with polypoid shape between the proximal and distal jejunum.
Fig 4. Small bowel resection was performed and the length of the resected bowel was 120 cm. In the resected specimen, there were 16 polypoid submucosal hematomas and the largest was 1.5 x 1.3 x 0.8 cm.

Fig 5.

A. The lesion consisted of ecstatic vasculature and organizing thrombi. (H&E stain ×200)

B. Elastic stains confirmed that the dilated and tortuous vasculature was composed of arteries and veins with direct communication. (Elastin stain ×200)
Fig 1