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CASE REPORT | SMALL BOWEL

Solitary Polypoid Angiodysplastic Lesion Mimicking a Tumor in the Jejunum

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ABSTRACT

Small bowel bleeding should be considered in patients with overt or occult gastrointestinal hemorrhage after normal upper and lower endoscopic examination. Angiodysplasia appearing as multiple flat vascular tufts is the most common cause of small bowel bleeding in patients over 40 years old. Polypoid angiodysplasia, however, is extremely rare. This report illustrates a unique case of solitary polypoid angiodysplasia in the jejunum of an adult with chronic kidney disease, who presented with an occult gastrointestinal bleed. The angiodysplasia mimicked tumorous growth, potentially indicative of malignant neoplasm. The patient underwent surgical resection and was histologically diagnosed as having angiodysplasia.

INTRODUCTION

Approximately 5–10% of all gastrointestinal (GI) bleeding occurs from the small bowel, so the small bowel should be considered in patients presenting with overt or occult GI hemorrhage after normal upper and lower endoscopic examination.[1] Angiodysplasia is the most common cause of small bowel GI bleeding, particularly in elderly patients, and is associated with various conditions, including chronic kidney disease (CKD), aortic stenosis, von Willebrand disease, and others.[1] Angiodysplasia appears as multiple flat vascular tufts, and its polypoid form is extremely rare.

CASE REPORT

A 76-year-old woman with end-stage renal disease presented with generalized weakness for a duration of 2 weeks. She reported having maintenance hemodialysis for 3 years. At presentation her vital signs were stable, and the exam was unremarkable. Laboratory tests revealed iron-deficiency anemia with a hemoglobin 8.1 g/dL, and a fecal occult blood test was positive. After transfusion of one unit of packed red blood cells, she underwent upper endoscopy and a colonoscopy, which failed to reveal the source of anemia. As her hemoglobin continued to decline, despite no evidence of active bleeding, she received 2 more units of packed red blood cells. Occult small bowel bleeding was suspected. Capsule endoscopy revealed a bleeding mass in the jejunum (Figure 1). A single-balloon enteroscopy revealed a friable mass 3 cm in diameter, which occupied <25% of the circumference of the distal jejunal crest (Figure 2). No other lesions were identified in rest of the small bowel. Multiple cold forceps biopsies were taken, and the location was tattooed.

The biopsy results were non-diagnostic, indicating a small intestine mucosa with mild chronic inflammation and hemorrhage. As her hemoglobin continued to drop, 2 additional units of packed red blood cells were transfused. An exploratory laparotomy with wide resection of small bowel (20 cm) revealed a tan-gray, friable mass lesion measuring 2.4 x 2 × 0.9 cm with an ulceration in its center. Microscopic examination confirmed that the lesion

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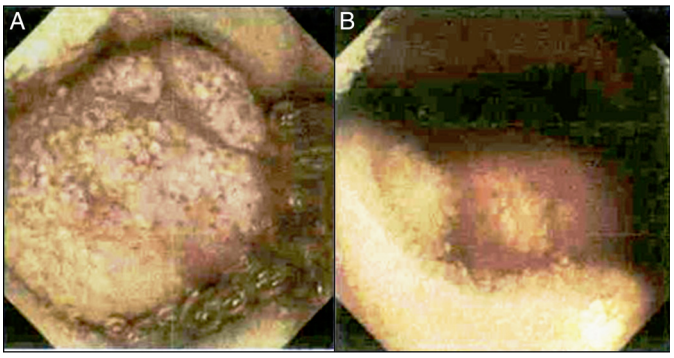

Figure 1. Capsule endoscopy of the small intestine demonstrating (A) a mass and (B) blood.

consisted of thin-walled, dilated blood vessels of various sizes, predominantly located within the submucosa (Figure 3). Thus the lesion was classified as an angiodysplasia, a benign vascular lesion. No other angiodysplastic lesions were identified in the resected specimen. The patient recovered fully from surgery. Upon routine follow-up 3 months later, she was asymptomatic, with hemoglobin 13.1 g/dL.

DISCUSSION

Angiodysplasia is an acquired, degenerative, non-neoplastic vascular lesion in the superficial layers of the bowel wall.[2] It is predominantly found in elderly patients and in patients with certain predisposing conditions, such as aortic stenosis, von Willebrand disease, and advanced CKD. Although the reason for increased prevalence of angiodysplasia in the advanced CKD population remains unexplained, it is likely that these lesions are not more common but are detected more frequently because of the increased risk of bleeding that is associated with uremia-induced platelet dysfunction.[3] However, the available evidence also indicates that an increased prevalence of angiodysplasia is directly related to the duration and severity of CKD,[4] which might be due to chronic intermittent fluid overload, leading to microvascular venous dilation and retrograde capillary dilation.[2] This mechanism differs from Heyde's syndrome, which is characterized as secondary to acquired von Willebrand factor deficiency.[5] However, most

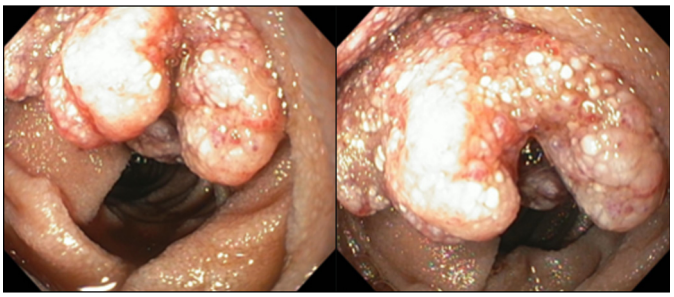

Figure 2. Enteroscopy demonstrating a 3-cm mass occupying <25% of the circumference of distal jejunal crest without oozing blood or surrounding venous dilatation.

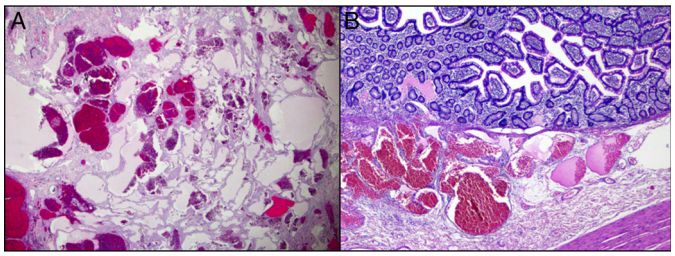

Figure 3. Hematoxylin and eosin stain of the resected specimen. (A) High-power magnification demonstrating a cluster of abnormally dilated, tortious blood vessels with thin walls and ectatic vasculature. (B) Low-power magnification demonstrating that these ectatic vessels are predominantly located in submucosa.

angiodysplasias are asymptomatic and incidental. If bleeding develops, it can be either overt or occult, but the majority of patients, including our patient, exhibit chronic and intermittent obscure GI bleeding, causing iron-deficiency anemia.[6]

On endoscopy, these lesions typically appear as a flat, bright red, dense reticular network of vessels measuring 2–8 mm in width. Patients often display multiple lesions, which tend to cluster in a particular segment of the GI tract. In contrast, the angiodysplasia in our patient appeared as a solitary polypoid lesion with bleeding, and it was thus suspected to be a neoplastic lesion. Polypoid angiodysplasia lesions in the GI tract are rare. A review of medical literature revealed only 18 recorded cases (excluding polypoidal arteriovenous malformations), 5 of which pertained to polypoid angiodysplasia lesions in the small intestine.[7] One of these 5 cases was a combination of hemangioma and angiodysplasia, while one was a pediatric case.[8,9] The remaining 3 cases were adults, of whom 2 had solitary lesions in the ileum and 1 had multiple bluish polypoid-shaped lesions in the jejunum, speculatively diagnosed as vascular lesions.[7,10,11] We believe that our patient's case is the first reported solitary polypoidal angiodysplasia lesion in the jejunum of an adult.

Similar to our case, endoscopic biopsy is generally of low diagnostic yield as significant histological abnormalities are submucosal in angiodysplasia.[12] Histologically, these lesions consisted of dilated, distorted, tortuous, thin-walled vessels that were lined by endothelium, with little or no smooth muscle and no inflammation, fibrosis, or atherosclerosis.[13] Although the terms "angiodysplasia" and "arteriovenous malformation" are used synonymously in pertinent literature and clinical practice, the two are histologically and pathophysiologically different. In contrast to angiodysplasia, arteriovenous malformations are arteriolized veins developed *in utero* with thick hypertrophic vessel walls.[13]

If an angiodysplasia is bleeding or has an overlying clot, treatment is indicated and endoscopic argon plasma coagulation is the preferred therapy. To establish a histologic diagnosis and remove the source of ongoing GI bleeding, we opted for

surgical resection instead of endoscopic treatment, which resulted in complete resolution. In a review of all reported cases, solitary polypoidal angiodysplasias in small intestine underwent surgical resection for histologic diagnosis and treatment, whereas polypoidal angiodysplasias in the large intestine was managed conservatively through endoscopic polypectomy, with complete resolution and hemostasis.[8,10,11]

The ability to diagnose these polypoidal angiodysplasias endoscopically and ascertain the level of risk associated with endoscopic removal of these solitary lesions in the small intestine can be challenging. With further research, managing these type of lesions in small intestine conservatively by endoscopic therapy may be possible.

DISCLOSURES

Author contributions: AD Polavarapu and M. Kesavan wrote and edited the manuscript. VV Gumaste and E. Purow critically revised the manuscript. M. Wrzolek supplied pathology images. AD Polavarapu is the article guarantor.

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