

CASE REPORT

Angiodysplasia as a Cause of Severe Hematochezia in a Child with End-Stage Renal Failure

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Abstract

Angiodysplasia is a frequent cause of gastrointestinal bleeding in adults with chronic renal failure (CRF); however, there is no data about this association in children. The mechanism of this association is not known. We report a 4.5-year-old boy with CRF presenting with hematochezia due to colonic angiodysplasia. He was on hemodialysis for the previous 9 months. Treatment with argon plasma coagulation (APC) was commenced following a short course of octreotide therapy. During the 3 years of follow-up, no occult or gross bleeding occurred. This case illustrates that octreotide and APC therapy seems to be useful for arresting bleeding from angiodysplasia and prevention of recurrent bleeding in children with CRF.

Keywords: angiodysplasia, end-stage renal failure, gastrointestinal bleeding, children, hemodialysis

INTRODUCTION

Angiodysplasia is characterized by dilatation, distortion, or thinning of blood vessels of the mucosa. The pathogenesis of angiodysplasia is still unclear. It can occur in various organs. In the gastrointestinal (GI) tract, the most common sites are the colon and distal ileum. It is recognized as a major casual factor of GI bleeding in the elderly population, but in younger adults, especially in children, diagnosis of angiodysplasia is extremely rare. In advanced chronic renal failure (CRF), unexplained GI bleeding (overt or occult) and anemia are common complications.¹ Angiodysplasia is a common cause of GI bleeding among adults with CRF^{2,3}; however, there is no data about this association in children. Herein, we report a boy with CRF who presented with massive hematochezia due to a colonic angiodysplasia.

CASE

A 4.5-year-old boy with CRF was admitted to our hospital with massive hematochezia. He was diagnosed

with focal segmental glomerulosclerosis at 5 months of age. Peritoneal dialysis was started when he was 3 years old. He was switched to hemodialysis 9 months ago due to recurrent peritonitis attacks. He was taking antihypertensive agents (amlodipine 0.6 mg/kg/day, propranolol 3.4 mg/kg/day, enalapril 0.6 mg/kg/day, doxazosin 1 mg/day), L-thyroxin, and digoxin at that time. During the 9-month period he was given erythrocyte transfusions every 2 weeks despite the absence of manifest bleeding and the usage of erythropoietin 150–200 units/kg/week. Physical examination revealed a blood pressure of 130/95 mmHg, heart rate of 128/min, pallor, and weakness. His weight and height were less than 3rd percentile for age and gender. Laboratory tests were as follows: Hb 6.7 g/dL, hematocrit 20%, MCV 78 fL, platelet count 320,000/mm³, white blood cell count 12,000/mm³, urea 130 mg/dL, and creatinine 5.7 mg/dL. Massive bleeding continued throughout the day (16 bloody stools/12 h) and multiple blood transfusions were given. Abdominal ultrasound showed dilated colonic segments with a high fluid content. Optimal

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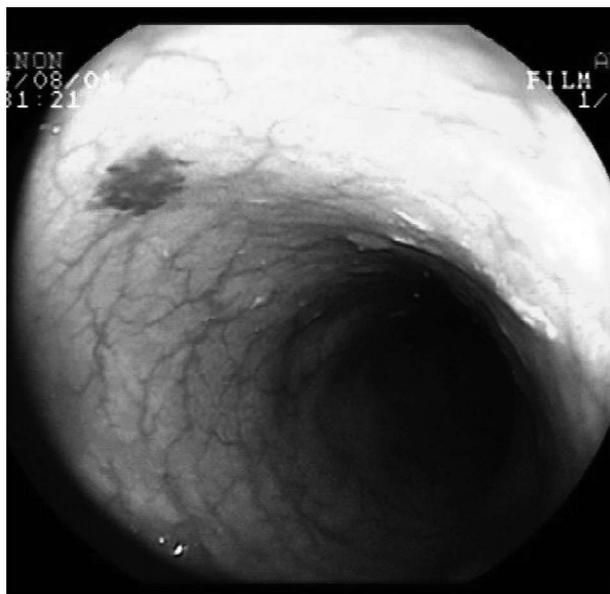


Figure 1. Endoscopic view of angiodysplasia in descending colon.

examination could not be performed by lower GI endoscopy because of massive bleeding. Meckel scintigraphy was found normal. Octreotide therapy (0.5 mcg/kg IV bolus, 0.5 mcg/kg/h IV continuous infusion) was started due to ongoing severe bleeding, and bleeding ceased during the third hour of infusion. Octreotide infusion was stopped after 12 h. Octreotide is well tolerated. Colonoscopy was performed on the second day, which revealed multiple angiodysplastic lesions in the descending colon (Figure 1). He was treated with argon plasma coagulation (APC) (40 W, 1.5 L/min flow). Short bursts (1–2 s) were used. No complication was seen. He had no rectal bleeding after APC, with a significant decrease of the need of blood transfusion and improvement of anemia. This favorable outcome was maintained during the 3 years of follow-up.

DISCUSSION

Prevalence of angiodysplasia as a cause of hemorrhage in adult CRF patients ranges from 19% to 47% compared to 5–17.6% patients with normal renal function.^{1,2} Angiodysplasia is the most frequent cause of recurrent bleeding in patients with CRF. However, the mechanism of this association is not known. To the best of our knowledge, this is the first pediatric case of angiodysplasia associated with CRF.

Microscopically, angiodysplasia consists of dilated, thin-walled, distorted vessels lined by endothelium and, infrequently, by a thin layer of smooth muscle. Submucosal veins and venules are primarily affected.³ The etiology of angiodysplasia is still unknown, and it could be related to vascular degeneration with aging accelerated by hypooxygenation of intestinal mucosa secondary to atherosclerotic peripheral vascular disease

or underlying disease such as aorta stenosis.³ The lesions of angiodysplasia may occur in any part of the intestine, most notably in cecum or ascending colon in adults. However, in children, the localization of angiodysplasia is reported to be in rectosigmoidal segment⁴ and the pathogenesis of this lesion in pediatric patients may be different.

Clinical findings range from unexplained iron deficiency to massive GI bleeding due to various degrees and variable intensity of bleeding.⁵ Hemorrhage is usually painless as in our case, ceases spontaneously in at least 90% of the cases, and recurs in 25–47% of them.⁶ Diagnosis of angiodysplasia is usually difficult and no gold standard as a diagnostic test is available. Colonoscopy and angiography are effective means of diagnosis; however, sensitivity of endoscopy is much higher compared to angiography (80% vs. 20%).⁷ Capsular endoscopy, double-balloon enteroscopy, and 99m Tc-labeled red blood cell scintigraphy can be used when endoscopy fails. Endoscopically, lesions appear flat or slightly raised above the mucosal surface, cherry red in color, and 2–10 mm in size. Lesions are multiple in 40–75% of cases as in our patient.⁶

Many therapeutic strategies have been used in patients with angiodysplasia such as endoscopic hemostatic therapy (EHT), surgical resection, and administration of estrogens, octreotide, thalidomide, tranexamic acid, epsilon amino caproic acid, or desmopressin.⁸ In adults with active hemorrhage, EHT is generally the initial form of treatment. In adults, there are numerous reports about endoscopic treatment of vascular lesions by APC⁹ whereas reports about APC used for treating vascular lesions in children are limited.¹⁰ Khan et al.¹⁰ reported that APC is an efficient and safe method for hemostasis and tissue ablation in pediatric cases. In this case, APC was successfully performed following a short course of octreotide therapy for acute bleeding without adverse effect. Somatostatin and its long-acting analogue, octreotide, have been suggested to be therapeutically helpful in bleeding angiodysplasia, in a systematic review of prospective observational studies.¹¹ In children with other vascular malformation of GI tract, anecdotal use of octreotide in doses ranging from 4 to 8 mcg/kg have been reported.⁸ However, in pediatric population, experience with octreotide therapy for angiodysplasia is very limited. The only published report on this topic was provided by Kaya et al.¹² They reported a 14-year-old boy who presented with upper GI bleeding due to large gastric angiodysplasia in whom octreotide 0.1 mg, subcutaneously, two times per day arrested bleeding within 2 weeks and the effect was maintained for the next 16 months of octreotide therapy. The rationale for the use of octreotide in the bleeding from angiodysplasia is based on its effects on splanchnic circulation. This drug induces a marked reduction of portal and mesenteric blood flow mediated through inhibition of vasodilator peptides.⁸ It has also anti-angiogenic activity in

vitro, which might also be relevant, because angiogenesis seems to play a role in the pathogenesis of angiodysplasia.¹³ Therefore, octreotide therapy was initiated in our patient. Octreotide is generally used in doses of 1 mcg/kg intravenous bolus followed by 1–2 mcg/kg/h continuous infusion for emergency treatment of GI bleeding in children.¹⁴ Clearance can be decreased by 50% in severe renal failure requiring dialysis. Although no details were found in the literature we adjusted the dose and gave half doses. The side effects of octreotide are generally mild such as abdominal pain, diarrhea, weakness, and skin rash. The more serious side effects are development of gallstones, hypothyroidism, kidney stones, and pancreatic enzyme deficiency.⁸ The relationship between octreotide therapy and cessation of bleeding is suggestive of a therapeutic effect in our case. We propose that octreotide may have a beneficial role in controlling massive GI bleeding due to angiodysplasia whenever endoscopic therapy is not promptly available or inapplicable.

We conclude that angiodysplasia may be a cause of acute or chronic rectal bleeding in children with CRF. Octreotide and APC therapy seems to be useful for arresting bleeding from angiodysplasia and prevention of recurrent bleeding in children with CRF.

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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