

## A Rare cause of Rectal and Colonic angiodysplasia: Culprit of Obscure Gastrointestinal Bleeding Masquerading as Radiation Proctitis

Richmond R Gomes

Professor, Medicine Ad-din Women's Medical College Hospital, Dhaka Bangladesh.

### Article Info

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**\*Corresponding author:** Richmond R Gomes, Professor, Medicine Ad-din Women's Medical College Hospital, Dhaka Bangladesh.

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### Abstract:

In 1974 was the first time that to describe abnormal distributions of the vessels of the submucosa of the colon, used the term angiodysplasia (AD). Angiodysplasia or vascular ectasia of the colon is a rare but important cause of recurrent lower gastro-intestinal bleeding and should be kept in mind as a diagnostic possibility. The aim of this paper is to report a case of simultaneous angiodysplasia of the colon and rectum. Here we present a 52 years old diabetic male who was previously diagnosed as a case of carcinoma prostate with completed 37 cycles of radiotherapy 2 months back. He presented with recurrent large volume hematochezia that necessitates repeated blood transfusion. Later he was started treatment as radiation proctitis with no improvement. Ultimately, he was diagnosed as sigmoid colonic and rectal angiodysplasia on third occasion of bleeding and given argon laser photocoagulation (APC) therapy. The patient recovered well without further symptom recurrence 5 months post discharge. We review the literature on colonic angiodysplasias and discuss the diagnostic challenges in obscure gastrointestinal bleeding.

**Key Words:** angiodysplasia; vascular ectasia; hematochezia; radiation proctitis; argon laser photocoagulation.

### Introduction:

Angiodysplasia or vascular ectasia or angioma of the colon is recognized as an important cause of lower gastro-intestinal bleeding in the elderly. [1] It usually involves the caecum and the right colon in adults. [1] The first case of angiodysplasia (AD) reported in the literature was described in 1839, but it was not until 1974 that the term AD was used to describe abnormal distributions in the submucosal vessels of the colon [2]. AD may be defined as the finding of abnormal, ectatic, dilated, winding and usually small (< 10 mm) blood vessels. Histologically, the vessels involved are exclusively limited to the endothelium with little or no presence of connective tissue [3]. Several publications suggest that the colon is one of the most frequent sites for the occurrence of AD [4,5] and angiodysplasia of the colon was reported for the first time as a hemangioma of the sigmoid colon by Holman [6], and Marguillis [7], et al. It may be asymptomatic and discovered incidentally during colonoscopy. [8] Patients may present with hematochezia, melena, positive occult blood test or iron deficiency anemia. [9] It may present as an isolated lesion or multiple vascular lesions. [10] The exact cause of vascular ectasia is not known but it is thought to occur due to ageing and degeneration of blood vessels. [11] An hypothesis states that it is due to contraction of the muscular layer leading to partial

occlusion of the sub-mucosal veins of the intestinal wall, subsequently the veins become tortuous and dilated. [11]

It predominantly affects the cecum and right side of the colon. [8,9] It could also affect any part of the large bowel. [10] Angiodysplasias have been reported in association with aortic stenosis, chronic renal failure, Von Willebrand's disease and cirrhosis of the liver. [12,13] The incidence of angiodysplasias is 0.8% in healthy people undergoing screening with colonoscopy in the US. [10,14] The incidence in other parts of the world is not known because of paucity of data.

There is no sex predilection. It is found predominantly in elderly people above the age of 65 years. [10] Diagnosis of angiodysplasias may be made using colonoscopy, angiography, and computed tomography (CT) scan and endoscopic forceps biopsy, which may reveal characteristic histopathologic features of dilated, distorted, thin-walled vessels, however they are not generally recommended because of low diagnostic yield and risk of bleeding. [15] Treatment could be conservative, medical, endoscopic or surgical treatment, but because of risk of re-bleeding surgical treatment is thought to be the best modality of treatment. [9] However, treatment should be individualized depending on severity. [9]

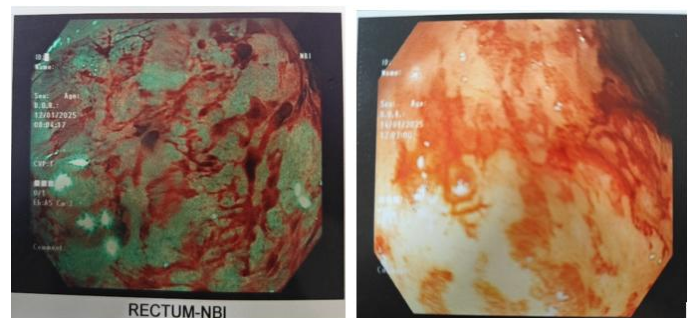
The purpose of this case is to report a patient who presented angiodysplasia of the colon and rectum simultaneously.

#### Case report:

A 52- year old diabetic gentleman, from Bangladesh presented with 2 months history of intermittent painless per rectal bleeding and symptoms of anemia. The character of bleeding was fresh to and mixed in the stools. He had previous history of hospitalization for two time in between and treated conservatively with repeated blood transfusion. His previous medical history was significant for carcinoma prostate with completed 37 cycles of radiotherapy 3 months back. During discharge he was advised to continue mesalazine. The patient had a total of two hospitalizations for bleeding within a span of 2 months. In total, 07 units of packed red blood cells were administered to his during this period. Hemoglobin level ranged from 5.0 g/dL to 9.3 g/dL. Peripheral blood film, platelet count, PT, aPTT was normal all through.

He was initially admitted from the emergency department for a history of fresh blood mixed in with stools for several days as well as symptomatic anemia 2months back. He underwent colonoscopy, which showed fresh blood streaks from the descending colon to rectum. No diverticulosis or neoplasia was found on colonoscopy. He was managed conservatively. Bleeding had resolved and stools were normal prior to discharge. He represented 3 weeks later for recurrent bleeding and anemia. Colonoscopy and in addition, gastroscopy was performed, which was normal up to the second part of the duodenum. Colonoscopy showed patches of erythema, erosions and vascular dilations all over rectum suggestive of radiation proctitis. This time he was treated with blood transfusion, hydrocortisone, metronidazole, sucralfate and mesalazine. Biopsy was not taken. As before, his symptoms resolved, and he was discharged well.

This time he again presented with fresh per rectal painless bleeding for 4 days and extreme fatigue for the same duration. He denied any abdominal pain, fever, abdominal mass, vomiting or weight loss. On examination, he was severely anemic, pulse 110 beats/min, regular with low volume, blood pressure was 80/60 mm of Hg. There was no abdominal tenderness, bowel sound was present. Complete blood count showed hemoglobin 4.9 gm%. He was resuscitated with intravenous isotonic fluid and 4 units of whole blood transfusion. Repeat full length colonoscopy was done which showed multiple vascular ectasia with evidence of recent bleeding seen in the rectum and sigmoid colon up to 25 cm from anal verge suggestive of colonic angiodysplasia. (Figure 1 and figure 2). Rest of the colon was normal. Biopsy was taken from rectum which revealed non-specific colitis. There were no histopathological features suggestive of ulcerative colitis. He was advised for APC 2 weeks later.



**Figure 1 and figure 2:** Colonoscopy showing multiple vascular ectasia with evidence of recent bleeding seen in the rectum and sigmoid colon respectively.

After 2 weeks APC was done (Figure 3 and figure 4.) The patient was observed in hospital for 48 hours post procedure, with no further bleed or drop in hemoglobin.



**Figure 3 and figure 4:** Colonoscopy showing normal rectum and sigmoid colon post APC.

He was discharged with 50 mg of thalidomide and sucralfate enema and found to remain symptom free at the outpatient clinic review 1 month later. There has been no recurrence of bleeding at 6 months post endoscopic therapy.

#### Discussion:

Aberrant blood vessels are most frequently found in the gastrointestinal tract where they are considered to be more common than at other sites. The terms “angiodysplasia”, “arteriovenous malformation”, “angioectasia” and “vascular ectasia” are synonymous. Angiodysplasias are differentiated from telangiectasias, which are similar but generally associated with congenital diseases. Additionally, some authors use the term “angioectasia” as a generic term, leaving “angiodysplasia” for lesions of the colon. [16] Vascular abnormalities can be divided into three large groups: a) vascular tumors or angiomas which may be benign (hemangiomas) or malignant (angiosarcomas); b) vascular abnormalities associated with congenital or systemic diseases, such as Klippel-Trenaunay Weber, Ehlers-Danlos, CREST and Osler-Weber-Rendu syndromes; and c) acquired abnormalities or sporadic lesions such as angiodysplasias [16].

Most ADs are detected in patients older than 60 years [12], although it has been reported in 30-year-old patients [17]. They have an 8% prevalence among healthy asymptomatic patients aged above 50 years old [12] and account for 3%-12% of acute lower GI bleeding. [18] Angiodysplasia has been associated with several systemic diseases, including end-stage renal failure, [19] and aortic stenosis, [20] none of which are present in our patient. GI bleeding from angiodysplasia in the presence of aortic stenosis is known as Heyde syndrome, after Edward Heyde who first described the

association in 1958. [21]

Their distribution is as follows: cecum 37%, ascending colon 17%, transverse colon 7%, descending colon 7%, sigmoid colon 18%, and rectum 14%. Western patients show involvement of the right colon (54 - 81.9%) [12], while the Japanese most frequently present involvement of the descending colon (41.7%) [4,22]. In our case, simultaneous presence in the right colon and rectum is remarkable, and it has not been reported in the literature.

The pathophysiology of angiodysplasia remains unclear. Proposed theories include chronic intermittent low- grade venous obstruction, resulting in dilatation of the submucosal veins and development of abnormal arteriovenous communications. [23] Other hypotheses include the involvement of angiogenic factors [24] as well as the association with aortic stenosis caused by a depletion of von Willebrand factor in blood flowing through a narrowed valvular stenosis. Laplace’s law explains this theory better, to evaluate wall tension, where tension is high when the radius is increased and wall thickness is decreased. This applies to the ascending colon and cecum (large diameter with thin wall) [25].

The sensitivity of colonoscopy for detecting angiodysplasia compared with angiography was found to be 81%. [26] Angiodysplasias are small, flat and cherry- red on endoscopic appearance, typically less than 10 mm in size. [12,27] Small lesion size, location behind a mucosal fold, or suboptimal bowel preparation during an emergent procedure may decrease the sensitivity of colonoscopic evaluation. Adding to the difficulty of detection is the natural history of angiodysplasia bleeding, which is typically slow, venous and intermittent. The low bleeding rate may also reduce the accuracy of multiphase CT angiography and formal angiography, which localize bleeding only if the rate of blood loss is greater than 0.3 to 0.5 mL/min and 0.5 to 1.0 mL/min, respectively. [27,28]

Radionuclide scanning is the most sensitive radiographic test for GI bleeding, able to detect bleeding greater than a 0.1 mL/ min. [29] After injection of the technetium- 99m pertechnetate- labelled red cells, a 60- minute dynamic scan is obtained, followed by static 2D planar images at intervals for several hours if no tracer accumulation is seen on the initial dynamic study. If bleeding is suspected on subsequent static imaging, an SPECT is performed for more accurate

localization. For these reasons, scintigraphy is useful for patients with obscure and intermittent bleeding, including those with suspected GI angiodysplasia. The 1999 AGA algorithm suggests a nuclear scan following upper and lower GI endoscopy for obscure overt GI bleeding. The disadvantage of a radionuclide scan is the inability to perform therapy, and a positive scan should be followed up shortly with angiography or colonoscopy for hemostasis. Delaying the therapeutic procedure may reduce the window for localization [30] and management of the lesion due to the intermittent nature of bleeding.

Various modalities have been used for treatment of bleeding angiodysplasias, including administration of octreotide or hormonal therapy, angiographic embolization, endoscopic methods or surgical resection. [18,31] Of the endoscopic techniques, argon plasma coagulation and bipolar or heater probe electrocoagulation have been best described in the literature, [18,32] with argon plasma coagulation recommended as first-line endoscopic therapy. [33] Mechanical hemostasis with endoscopic clips has not been well reported. Takahashi et al previously documented a case of successful endoscopic clipping for bleeding from colonic angiodysplasia in a case of Heyde syndrome. Aortic valve replacement has been shown to improve angiodysplastic bleeding in patients with aortic stenosis, [20] although the angiodysplasia has been reported to persist months later. [20,34] Our case underwent APC therapy.

A 2014 meta-analysis of 831 patients with angiodysplasia, including 623 patients treated via endoscopy, showed a 36% rebleeding rate at a median of 22 months from endoscopic therapy. [31] It may, therefore, be important to follow-up these patients over the longer-term.

Surgery is the definitive treatment for clearly identified lesions, aimed at patients with severe gastrointestinal hemorrhaging which cannot be controlled by any of the other therapies described above, or for patients with recurring chronic hemorrhaging, or dependent on blood transfusions [35]. We decided that our initial option was surgery after identifying the site of the lesion with no other concomitant disease, because we had a young, stable patient, and we expected good results for a very delicate pathology, with previous pathology report of angiodysplasia. Hormone therapy has also been described for treatment of AD, such as the use of

thalidomide which is an angiogenesis inhibitor that suppresses the vascular endothelial growth factor. Good results have been obtained in some series, but with a high number of adverse events, such as fatigue (32%), constipation (25%), dizziness (21%), and peripheral edema (14%), and less frequently, abdominal distention, thrombocytopenia, leukopenia, blurry vision, dry eyes, pruritus, rash, headache and herpes zoster [35].

#### Conclusion:

Colonic vascular ectasia may be asymptomatic or cause life threatening lower GI bleeding. Therefore, treatment should be individualized. In conclusion, this report highlights that angiodysplasia is a rare but important cause of recurrent intestinal bleeding in adult and should be kept in mind as a diagnostic possibility. Early diagnosis of this rare lesion is important to avoid a possible fatal outcome and thus the physicians should be aware of this lesion as a rare cause of intestinal bleeding in adult.

**Conflict of interest:** None declared

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