Hemangiomas of the Pelvis

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ABSTRACT

Hemangiomas of the colon and rectum are rare but often misdiagnosed causes of hematochezia. They are characterized by the clinical triad of recurrent episodes of painless rectal bleeding, multiple ectopic phleboliths on plain radiographs, and cutaneous hemangiomas. The majority of these tumors involve the rectosigmoid region. Diffuse cavernous lesions are the most common histological type. Endoscopy is important to define the extent and number of lesions. Imaging by computed tomography or magnetic resonance imaging is used to define invasion into pelvic structures. Surgical resection with preservation of the sphincters is the recommended treatment.

KEYWORDS: Hemangioma, colon, rectum

Objectives: Upon completion of this article, the reader should be able to summarize the diagnosis and management of colorectal hemangiomas.

INCIDENCE

Hemangiomas are benign tumors made up of mature well-formed vessels usually lined by a single layer of endothelium.¹ They are among the rarest tumors of the lower gastrointestinal (GI) tract. Phillips² described the first such case in 1839, of an 18-year-old man who presented with hemorrhage from an ulcerated anal hemangioma. Suture ligation of the lesion led to sloughing of the tumor and resolution of symptoms. Of the 255 cases of gastrointestinal hemangiomas Gentry et al³ identified in their exhaustive 1949 review of the literature and the Mayo experience, only 96 involved the colon, rectum, or anus. This number had increased to about 300 in the review by Coppa et al⁴ in 1984. Fifty percent of large intestinal hemangiomas are rectal in location.^{5,6}

Intestinal hemangiomas are important because of their potential for causing massive hemorrhage.^{3,7-11} Early reports cite a 40 to 50% mortality in untreated

cases.^{3,12,13} Death from exsanguination has been described.^{7,14} These tumors are difficult to diagnose, and rectal lesions in particular are frequently misdiagnosed and mismanaged.¹⁵ Indeed, in reviewing 47 pediatric cases of intestinal hemangiomas, Nader and Margolin¹⁶ found that the average time between onset of symptoms and correct diagnosis was 16 years. Jeffery et al¹⁷ found that 80% of patients who were finally diagnosed with a colorectal hemangioma had undergone a minimum of one inappropriate surgical procedure as a result of incorrect diagnosis. The triad of intermittent hematochezia, multiple ectopic phleboliths, and cutaneous hemangiomas should make the physician suspicious of a hemangioma as the cause of GI hemorrhage.¹⁸

DEMOGRAPHICS

Although intestinal hemangiomas overall are about 2.5 times more common in men than women, this

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Table 1 Cutaneous Syndromes Associated with Gastrointestinal Hemangiomas

Syndrome	Inheritance	Characteristics
Blue rubber bleb nevus syndrome (BRBNS)	Most cases are sporadic, but families with autosomal dominant have been described. Putative genetic defect on chromosome 9.	Cavernous hemangiomas of the skin, gastrointestinal tract, other viscera. Cutaneous lesions are blue, tender lesions 0.5–5 cm in diameter—with compression, blood can be emptied out of them. A recent case report has suggested that pregnancy may induce intestinal hemangiomas to bleed.
Klippel-Trenaunay-Weber syndrome	Sporadic	Triad of cutaneous hemangiomas, bone and soft-tissue hypertrophy of the lower extremities, and congenital varicose veins.
HHT (Osler-Rendu-Weber syndrome) (Nader and Margolin 1966) ¹⁶	Autosomal dominant	Mucocutaneous telangiectasia especially oral, nasal. Hemangiomatous lesions have been found in the stomach, small intestine, and rectum.

does not seem to be the case specifically for colorectal hemangiomas, for which there is a roughly equal sex distribution.³ A significant proportion of patients are symptomatic from childhood, but the diagnosis is often not made until adulthood. Thus, there is a wide age range at time of definitive diagnosis, from 2 months of age up to 79 years.^{3,5,19}

ASSOCIATIONS

Hemangiomas of the colon and rectum can occur as solitary or multiple lesions. There may be synchronous lesions of the stomach and small intestine. They may be a part of a multicentric process with involvement of the spleen, kidney, brain, and skin.^{20,21}

The association of cutaneous lesions with GI hemangiomas was first recognized by Gascoyen²² in 1860. Since then, several syndromes have been described, including blue rubber bleb nevus syndrome (BRBNS), Klippel-Trenaunay-Weber syndrome, and Peutz-Jeghers syndrome (see Table 1). However, cutaneous hemangiomas are rarely (1.8% of cases) associated with intestinal visceral hemangiomas.²³

There is a well-established connection between the use of conjugated estrogens, as found in the oral contraceptive pill, and the growth of hepatic hemangiomas. No such association has been described for intestinal hemangiomas.

CLASSIFICATION

Building on the work of Kaijser,²⁴ intestinal hemangiomas have been divided into several categories^{3,18,20,25,26}:

- Capillary
- Cavernous
 - Localized (polypoid or nonpolypoid)

Diffuse infiltrating (expansive)

Multiple phlebectasia

- Mixed
- Hemangiomatosis

MICROSCOPIC AND MACROSCOPIC PATHOLOGY

Almost all intestinal hemangiomas are polypoid and intraluminal; the occasional solitary tumor may be annular, encircling the bowel wall. They range in color from red to purple and are soft and compressible unless they are thrombosed and contain phleboliths. The normally pink-tan mucosa adjacent to the tumor is often congested and edematous.³

Capillary hemangiomas constitute 5 to 10% of benign vascular tumors of the GI tract.^{3,6,27} They consist of small, thin-walled, densely packed vessels located in and originating from the submucosal vascular plexus. Vessels are lined with a single layer of well-differentiated endothelium. There is minimal surrounding connective tissue, especially with regard to smooth muscle and elastin. Capillary hemangiomas are more common in the small intestine, appendix, and perianal skin. They are usually solitary and, despite appearing well circumscribed, lack a true capsule. Mucosal ulceration is present in 50%. Edema and chronic inflammatory changes in the overlying mucosa can make diagnosis difficult. The lining endothelial cells are immunoreactive with antibodies to factor VIII-related antigen.⁶

Cavernous hemangiomas, although similarly located in the submucosa, consist of larger vessels with a predilection for forming blood-filled sinuses. There may be a single or multiple layers of flattened endothelial cells, with scant interstitial tissue.²⁸ Thrombosis within these tumors is common. They constitute one third of all benign vascular tumors of the GI tract.²⁹ Three subtypes

Figure 1 Cavernous hemangioma. Photomicrograph showing the dilated, congested submucosal vessels characteristic of this type of hemangioma. (Courtesy of Dr. J. Ritter, St. Louis, MO.)

have been described. Localized cavernous hemangiomas, when polypoid, are usually sufficiently large to be symptomatic. The diffuse expansive type (see Fig. 1) typically extends along some 20 to 30 cm of intestine and may be multiple. The rectum is involved in 70% of cases,³ in which there may be involvement of the entire thickness of the intestinal wall and infiltration into surrounding structures.^{4,6} The term "multiple phlebectasia" refers to the presence of multiple discrete tumors less than 1 cm in diameter.

Mixed hemangiomas constitute 5 to 10% of benign intestinal vascular tumors. They share variable features of both capillary and cavernous hemangiomas and have a distribution similar to that of the former. They are typically polypoid.⁶

Disseminated intestinal hemangiomatosis refers to the presence of more than 50 intestinal hemangiomas spread throughout the GI tract. Histologically, they are usually of the cavernous type. These lesions are part of a syndrome of disseminated hemangiomatosis affecting three or more systems. Patients typically present at birth with numerous cutaneous hemangiomas.⁶

PATHOPHYSIOLOGY

Colorectal hemangiomas are congenital, arising from embryonic sequestrations of mesodermal tissue. They enlarge by the proliferation of their endothelial cells, impinging upon the overlying mucosa as well as the muscularis beneath. The different histological types reflect anomalies occurring at different stages of stem cell development—capillary hemangiomas at the initial stage, cavernous at the second stage.¹⁸

The vascular channels of hemangiomas, probably through a combination of disordered blood flow and abnormal endothelium, initiate the coagulation cascade. This leads to platelet sequestration,³⁰ systemic

thrombocytopenia, and a coagulopathy similar to disseminated intravascular coagulopathy (DIC), with depletion of fibrinogen and factors V and VIII.^{4,31} The triad of a rapidly enlarging hemangioma, thrombocytopenia, and coagulopathy is known as Kasabach-Merritt syndrome. Despite being extremely rare, it is associated with a 30 to 40% mortality rate related to uncontrollable hemorrhage.²⁰ Management involves stabilization of hemostasis by replacing consumed clotting factors and platelets while trying to remove or ablate the lesion. For the latter, options described include angiographic embolization, steroids, interferon- α , and chemotherapy. Use of anticoagulants, as in DIC, remains controversial.³²

Local thrombus formation may fragment reticulocytes, which may lead to a microangiopathic hemolytic anemia. Microvascular obstruction of the submucosa and overlying mucosa may cause segmental ischemia and secondary inflammatory changes.³¹

Numerous reports have described invasion of the hemangioma into surrounding structures, particularly the bladder, uterus, and sacrum.^{11,33–35} Gentry et al³ also reported two patients who developed rectovaginal fistulas.

Erosion or ulceration into the abnormal vessels that constitute the hemangioma leads to profuse bleeding because of the lack of muscular and supportive elastic tissue in the walls.

Malignant transformation is exceedingly rare.²¹

CLINICAL FEATURES

History

Eighty percent of hemangiomas are symptomatic. Their outstanding clinical feature is intraluminal hemorrhage.^{6,36} Recurrent episodes of painless bleeding are characteristic and occur in 60 to 90% of cases. Episodes begin in childhood and are recurrent throughout adolescence, becoming progressively more severe.⁴ Most reported cases present with large-volume hematochezia. More than 50% of patients demonstrate evidence of chronic, iron deficiency anemia.²⁵

In general, the cavernous type tend to bleed massively much more frequently than the capillary type because of their greater size and larger blood volume. Distal (i.e., rectal) lesions are more frequently symptomatic than more proximal lesions.

Bleeding occurs because of erosion of the walls of the abnormal vasculature through the mucosa into the intestinal lumen. As stool moves distally through the colon it solidifies, which can lead to greater trauma to the mucosal surface. This helps to explain why distal lesions are more likely to bleed than right colon or small intestinal lesions. However, in rare cases of transmural hemangiomas, rupture can occur on the serosal surface, leading to retroperitoneal or intraperitoneal hemorrhage.

It is important to note that the size of a hemangioma does not always correlate with the amount or rate of bleeding. Condon and Loyd³⁷ described a patient with massive hematochezia from a single 1.8-cm sessile nodule in the mid-descending colon.

Colorectal hemangiomas infrequently cause large bowel obstruction.^{38,39} A diffuse tumor may completely encircle the bowel wall, leading to luminal stenosis and obstruction. Polypoid lesions may rarely act as the lead point for intussusception.^{3,40} Large tumors of the rectum may cause constipation with tenesmus; defecation may exacerbate the bleeding.⁶

Allred and Spencer,²⁵ in a case series of 29 patients from the Mayo experience, noted that 14% of patients had diarrhea.

Physical Examination

The presence of cutaneous hemangiomas (see earlier) may be an important clue to intestinal hemangioma being the cause of GI bleeding.

A large colonic hemangioma may be palpable as an abdominal mass. Similarly, a rectal hemangioma may be palpated on digital rectal examination as a soft ("doughy"), smooth, nodular, compressible mass that cannot be well outlined by the palpating finger.⁴¹

INVESTIGATIONS

Laboratory Studies

As discussed previously, abnormal findings may include anemia (of both the iron deficiency and acute blood loss varieties), thrombocytopenia, hypofibrinogenemia, coagulopathy, and decreased levels of factors V and VIII.

Imaging

Hollingsworth¹⁵ was the first to report the association of a narrowing of the sigmoid colon on barium enema with an area of surrounding phleboliths in a 9-year-old girl who had previously been operated on for peripheral hemangiomas.

PLAIN ABDOMINAL FILMS

Phleboliths are calcified, well-circumscribed densities (usually multiple) particularly associated with colonic cavernous hemangiomas. They are present in up to 50% of cases and do not reliably indicate the extent, nature, or number of the hemangiomas present.³¹ This is because development of radiographically visible phleboliths is a gradual process, and thrombosis, which leads to phlebolith formation, is primarily limited to the portion of the tumor within the colonic wall, so that soft tissue extension is not visualized. One should be suspicious that pelvic phleboliths may reflect the presence of a

hemangioma when observed to be clustered in atypical locations (more caudal and medial) in a young patient.^{4,42} In contrast, less than 5% of normal subjects show any radiographic evidence of pelvic phleboliths prior to the third decade.¹⁴ Lateral radiography distinguishes phleboliths in the presacral space from the more common anterior phleboliths in the pelvic venous plexus.^{5,20} Large hemangiomas of the rectum may cause displacement or distortion of the rectal air column seen on plain films.⁴³

BARIUM ENEMA

Barium contrast studies are most useful for identifying polypoid or obstructing lesions. Features characteristic of a hemangioma on contrast enema are:

- Presence of a luminal filling defect, especially "scalloping" of the rectal wall produced by nodular elevations of hemangiomatous tissue⁴²
- Narrowing of the distal rectosigmoid^{4,43}
- Widening of the prerectal space as a mass effect of the lesion⁴³

Displacement of small bowel loops may provide an indication of the true dimensions of the tumor. The more rigid the affected bowel loops are, the older the lesions are, as a consequence of chronic inflammation with scarring of the bowel wall.⁵

COMPUTED TOMOGRAPHY SCAN

Although CT can be useful in confirming the presence of the tumor in the form of heterogeneous thickening of the bowel wall and engorged vessels in the perirectal fat, it has a more important role in defining the extent of the lesion and the presence of invasion into the mesentery or surrounding structures.³³

SELECTIVE MESENTERIC ANGIOGRAPHY

Selective mesenteric angiography can detect the hemangioma even in the absence of active bleeding. The vascular malformation takes the form of venous pooling or "puddling," most commonly in the rectosigmoid region. It is also very useful for detecting synchronous lesions.⁵ Delayed-phase studies assist in demonstrating large venous malformations.⁴ However, thrombosis within these tumors may cause them to appear hypovascular or avascular, thus obscuring the true extent of the lesion.¹⁰

MAGNETIC RESONANCE IMAGING

MRI (Fig. 2) provides detailed anatomic images as well as being able to demonstrate blood flow anomalies without the need for contrast medium.⁴⁴ T2-weighted images (more clearly depicted with fat suppression) show a markedly thickened rectosigmoid wall with very high signal intensity.^{19,45,46} Thrombosed vessels within the

Figure 2 T2-weighted magnetic resonance image of the pelvis demonstrating increased signal intensity of the submucosal layer, infiltrating through the bowel wall and throughout the perirectal fat, consistent with rectal hemangioma. (Courtesy of Dr. C. Menias, St. Louis, MO.)

tumor mass appear as tubular structures with high signal intensity. Addition of an endorectal surface coil, as in endorectal ultrasonography, is able to evaluate the extent of tumor invasion through the five separate layers of the bowel wall and the sphincter muscles.⁴⁴

ENDOSCOPY

Colonoscopy is essential in the evaluation of these patients. Hemangiomas appear as submucosal projections, ranging from a deep blue to a dull red ("plum" colored).^{6,43} Although frank ulceration is usually not visible, there may be several pinpoint bleeding areas. The tumors collapse on insufflation.³¹ The overlying rectal mucosa is edematous, suggesting chronic inflammation, which may lead the endoscopist to form the alternative (but erroneous) diagnosis of inflammatory bowel disease.^{3,17,20,31}

The proximal extent of the lesion should be determined. In the stable patient with no active bleeding, a complete survey of the colon and the upper GI tract is important to exclude synchronous lesions. Biopsy may cause severe hemorrhage and is usually contraindicated.^{4,20,25,31,42,47} Nevertheless, Wang⁴⁸ has suggested that biopsy of a carefully selected area may be useful in confirming the diagnosis.

TREATMENTS

Operative resection remains the recommended treatment, whenever possible, for colorectal hemangiomas because of the high risk of recurrent bleeding. Non-operative therapy (as described in the following) is limited, for the most part, to case reports with little long-term follow-up.

Local control of bleeding (e.g., hemorrhoidectomy) has had little success.¹⁷ Early conservative measures were complicated by rebleeding. Gabriel^{49,50}

reported success with ligation of the arteries feeding the lesion, but this technique appears unreliable as subsequent bleeding has occurred in other patients.¹² Bancroft¹³ ligated the superior hemorrhoidal vein and injected it distally with 10 mL of 40% sodium salicylate. Despite initial success, the hemangioma recurred over the course of 20 years and ultimately required abdominoperineal resection.⁵¹

Pharmacological Therapy

To date, there are no known pharmacological agents that have been shown to be efficacious for intestinal hemangiomas. Steroids have been used in the treatment of infantile cutaneous hemangiomas, with a response rate of 30 to 60%.^{52,53} Interferon- α 2a has been shown to be effective in inducing regression of life-threatening, steroid-resistant hemangiomas of infancy, but none of the 20 patients in this study had known intestinal lesions.⁵⁴

Radiotherapy

Irradiation of vascular tissue induces proliferation of the intima and fibrous sclerosis of the walls, sometimes with fibrinoid changes and occlusion by fibrin thrombi and development of arteriosclerosis.⁵⁵ Radiation therapy has been successful in controlling bleeding from rectal tumors^{41,55,56} but would not appear to be appropriate for nonrectal lesions because of the toxicity to the adjacent organs.

Endoscopic Therapy

Polypoid tumors with a narrow base can be removed by snare polypectomy with electrocautery.⁵⁷⁻⁵⁹ However, such patients should be observed closely for postprocedure bleeding.⁶⁰ Azizkhan⁹ described a patient with a diffuse rectosigmoid hemangioma who, after failing excision and oversewing, then sclerotherapy, was having daily massive episodes of hematochezia by age 17. Argon laser therapy resulted in almost complete remission of his bleeding. Despite these successes, both patient and endoscopist must be aware of the risk of inducing uncontrollable hemorrhage and the need for emergent laparotomy if this occurs.

OPERATIVE MANAGEMENT

Choice of operation for patients with rectosigmoid lesions includes limited colon resection, low anterior proctosigmoidectomy with colorectal anastomosis, modified Parks coloanal pull-through, or abdominoperineal resection with end colostomy. Up until 1976, abdominoperineal resection was the procedure of choice for the resection of hemangiomas involving the distal rectum.^{4,18,20,51,61,62}

Since then, sphincter preservation using rectal mucosectomy and pull-through coloanal sleeve anastomosis has become the first-line procedure in these usually young patients with benign disease. After the first ileoanal anastomosis after rectal resection was described in 1947 by Ravitch and Sabiston,⁶³ Soave in 1964⁶⁴ refined this technique in children with Hirschsprung's disease to include rectal resection with mucosectomy of the retained muscular rectal cuff and a coloanal pull-through anastomosis. This was further modified by Parks⁶⁵ for the removal of villous adenomas of the rectum. The concept is to remove the hemangiomatic colon but to leave the abnormal rectum in situ. It is important to obtain early control of the inferior mesenteric artery.³¹ Limited pelvic dissection lessens the likelihood of severe intraoperative bleeding, hematoma formation, and damage to hypogastric, lumbar, and sacral nerves.⁶⁶ A surgical plane seems to persist between the mucosa and the underlying muscularis, which permits successful removal of the rectal mucosal sleeve.⁶⁷ The normal distal colon is then drawn down through the remaining muscularis cuff. The hemangiomatic vessels remaining in the rectal wall are no longer subjected to any local trauma, consequently reducing the risk of further bleeding.¹⁷ Cunningham et al⁴⁷ recommended leaving a short (3 to 4 cm) rectal cuff to minimize the amount of retained hemangioma.

In 1994, Londono-Schimmer et al⁶⁶ reported the long-term results of 15 patients treated using this method. There were no perioperative deaths, and complications included one leak with abscess formation and two perianastomotic hematomas. Three patients were lost to follow-up. Of the remainder, three patients had rare sporadic bleeding after wiping, one had a rectal cuff abscess, and another had occasional incontinence.

Management of Hemangiomatosis

Operative management of hemangiomatosis is confounded by the large number of lesions distributed throughout the GI tract, all of which are at high risk of ulceration and hemorrhage. In the actively bleeding patient, angiography and tagged red cell scans may assist in localizing the specific source lesion. Definitive management remains poorly defined. Wong and Lau⁶⁸ recommended an aggressive surgical approach in describing a patient with BRBNS who, in a single operation, underwent excision of a total of 69 hemangiomas—3 gastric, 1 duodenal, 59 jejunoileal, 3 colonic, 1 rectal, and 2 on the surface of the liver. This was accomplished through a combination of segmental resections, wedge resections, right hemicolectomy, and simple excisions through colotomies. The patient had an uneventful postoperative course and was discharged home on postoperative day 9. At 18 month follow-up, he was well with no recurrent bleeding.

There are several case reports describing endoscopic management of hemangiomatosis. Endoscopic laser photocoagulation has been reported as successfully treating numerous widespread lesions in patients with BRBNS.^{69–71} Bak et al⁷² used the snare polypectomy technique to remove some 18 colonic lesions over three sessions. Five lesions in the second part of the duodenum were also removed, and eight gastric lesions were banded. There were no complications, with no evidence of recurrent tumors or bleeding on repeated upper and lower endoscopies at 2 months. Shimada et al⁷³ took this approach further in another patient with BRBNS-associated hemangiomatosis. After endoscopic ablation of three gastric and seven colonic hemangiomas by electro-surgical snare, this patient underwent a laparotomy in which two enterotomies enabled endoscopic snare excision of 39 small intestinal cavernous hemangiomas. One year after an uncomplicated recovery, a combination of panendoscopy and small bowel contrast follow-through examination revealed no new or remaining lesions.

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