GI-Associated Hemangiomas and Vascular Malformations

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ABSTRACT

Hemangiomas and vascular malformations of the gastrointestinal tract, rare clinical entities, present as overt or occult bleeding. They can be distributed throughout the intestinal digestive system, or present as a singular cavernous hemangioma or malformation, which is often located in the rectosigmoid region. Misdiagnosis is common despite characteristic radiographic features such as radiolucent phleboliths on plain film imaging and a purplish nodule on endoscopy. Adjunctive imaging such as computed tomography and magnetic resonance imaging are suggested as there is potential for local invasion. Endorectal ultrasound with Doppler has also been found to be useful in some instances. Surgical resection is the mainstay of treatment, with an emphasis on sphincter preservation. Nonsurgical endoscopic treatment with banding and sclerotherapy has been reported with success, especially in instances where an extensive resection is not feasible.

KEYWORDS: Hemangioma, cavernous hemangioma, vascular malformation, blue rubber bleb

Objectives: Upon completion of this article, the reader should be able to summarize the characteristics, workup, diagnosis, and treatment of gastrointestinal hemangiomas and vascular malformations.

EPIDEMIOLOGY

First documented in 1839, hemangiomas and vascular malformations of the gastrointestinal (GI) tract are infrequently encountered entities.^{1,2} They may occur anywhere along the intestinal system; the small bowel is the most frequent site with hemangiomas and malformations accounting for 10% of all small bowel tumors.³ Colonic and anorectal hemangiomas and malformations are even rarer yet, with 200 cases documented from 1931 to 1974.^{4,5} A 1949 review of GI hemangiomas and malformations only found 38% located in the colon and rectum.² Another review classified 50% of colonic

hemangiomas and malformations being distally located in the rectum.⁶ To date, there have been over 130 reports of rectal hemangiomas/malformations.

With many hemangiomas and vascular malformations present at birth and often misdiagnosed, the age at which a definitive diagnosis is made has ranged from 2 months to 79 years.^{2,7,8} Generally, hemangiomas/malformations affect the young, with a male:female ratio of 1:2.5²; however colonic hemangiomas/malformations approach an equal male:female ratio. The largest operative single institution series to date ($n=10$), which looked at a subclassification of vascular malformations

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Table 1 Hemangiomas versus Vascular Malformations¹¹

	Hemangiomas	Vascular Malformation
Histology	High endothelial cell turnover	Normal endothelial cell turnover
Presence at birth	Usually absent	Present (not always apparent)
Clinical	Apparent 6–8 weeks after birth. Proliferative phase for 1–2 years then spontaneous involution	Grows in proportion with person
Diagnosis	Clinical history, appearance	Imaging (MRI, CT, US, angiography)
Treatment	Observation If not fully involuted, minor surgical correction. If large or in anatomically sensitive area, steroids or interferon gamma or surgical correction	Depending on site, size, symptoms, etc. From conservative only to laser for capillary malformations, sclerotherapy with or without excision, or surgery alone

MRI, magnetic resonance imaging; CT, computed tomography; US, ultrasound.

known as blue rubber bleb nevus syndrome had a mean patient age of 16 years old and a range of 2 to 36 years old.⁹ Initial bleeding symptoms were present early (mean age = 5 years).

HISTOLOGY, PATHOLOGY, AND CLASSIFICATION

The term “hemangioma” is often misused; this is further complicated by the confusing nomenclature.¹⁰ Adjectives and modifiers such as “strawberry,” “cavernous,” and “capillary” are added to the term “hemangioma,” but in the strictest sense are not hemangiomas.¹¹

In 1982, Mulliken and Glowacki classified vascular lesions in a consistent and meaningful scheme based on the histology and the endothelial cell turnover.^{12,13} High endothelial cell turnover entities are accurately termed hemangiomas (infantile hemangioma, rapidly involuting congenital hemangioma, noninvoluting congenital hemangioma, Kaposiform hemangioendothelioma, and tufted angioma). These lesions are present at birth and the majority undergoes a spontaneous involution. Administering steroids or interferon can accelerate this process. Normal endothelial conditions are correctly termed vascular malformations, classified by their dominant abnormality (arteriovenous malformation, venous malformation, lymphatic malformation, lymphatic-venous malformation, and capillary malformation).^{11,14} In 1996, the International Society for the Study of Vascular Anomalies approved this classification system (Table 1).

Admittedly, here the term hemangioma has been used because of clinical familiarity with the term. Although hemangiomas do occur in the GI tract, the far more dominant entities are vascular malformations, including “cavernous hemangiomas.” Henceforth the term “cavernous vascular malformation” or “cavernous malformation” will be used.

Vascular malformations result from an embryologic error in morphogenesis.¹⁴ Mature endothelial channels lack smooth muscle, allowing expansion over

time from hydrostatic means, and not proliferative expansion as hemangiomas do. A classification system of intestinal malformations has been created based on the histologic abnormality (Table 2).^{15–19}

The capillary subtype of malformations is located in the perianal skin, small bowel, and appendix.²⁰ Usually singular, they lack a true capsule and are well circumscribed: half can have associated mucosal ulceration, with accompanied edema and inflammation. The histologic hallmark is that they are a proliferation of capillaries with thin-walled spaces lined by endothelial cells.

Eighty percent of rectosigmoid malformations are the cavernous subtype.^{16–18,21,22} As opposed to the capillary malformations, cavernous malformations are large spaces lined by single or multiple layers of endothelial cells (Fig. 1). The localized variety of cavernous malformations is often polypoid and can be symptomatic. The diffuse variety of cavernous malformations has been reported up to 30 cm in length, and can be multiple. These are the entities that can be circumferential, and have the potential for local invasion to adjacent structures, with the rectum involved in 70% of instances.^{2,23}

The gross appearance of GI vascular malformations overwhelmingly present as intraluminal lesions, though diffuse cavernous malformations can extend into adjacent structures by infiltrating the submucosa and beyond. They range from solitary lesions to clusters.

Though GI vascular malformations can be isolated as separate entities, there are several associated syndromes with characteristic organ involvement. The

Table 2 Intestinal Vascular Malformation Classification^{15–19}

Capillary
Cavernous
Localized (polypoid or non-polypoid)
Diffuse infiltrating (expansive)
Mixed
Hemangiomatosis

Figure 1 Cavernous vascular malformation. It is composed of blood-filled sinus-like spaces with prominent vascular channels in the submucosa. (Left: hematoxylin & eosin [H&E] 4x; right: H&E 10x.)

presence of simultaneous vascular malformations in the skin, brain, spinal cord, along with other clinical traits such as limb and bone hypertrophy, varicose veins, and arteriovenous fistulas have been documented (Table 3).^{8,16,17,20,24} Despite cutaneous involvement being a frequent association, only 1.8% of cutaneous hemangiomas and vascular malformations have GI vascular malformations present.²⁵ Research in analyzing mono- and dizygotic twins found no evidence of predisposing inherited patterns.²⁶ This is however countered by a description of six families that demonstrated an autosomal dominant morphology with incomplete penetrance.²⁷

PATHOPHYSIOLOGY

In the original classification by Mulliken and Glowacki, the key differentiation between hemangiomas and vascular malformations was due to the smooth muscle proliferation.^{12,13} The abnormality arises in the embryologic mesoderm, related to a defect intrinsic to the endothelial cells and the secretion of growth factors. Hormonal influence is also believed to play a part in their development as evidenced by the 3:1 F:M ratio.² The developmental stage of when the angiogenic abnormality occurs accounts for the different histologic subtypes, with capillary malformation developing from an earlier defect than cavernous malformation in the stem cell cycle.^{2,8,16,28}

A sequestration of platelet and clotting factors occurs due to the alteration of blood flow and from the abnormal endothelium, initiating coagulation.²⁹ Kasabach-Merritt syndrome can develop in rapidly expanding hemangiomas, where a DIC- (disseminated intravascular coagulation) like picture can develop with consumption of fibrinogen and factors V and VIII, uncontrolled bleeding, and a 35% mortality rate.³⁰ The constant sequestration of flow can lead to calcification and phlebolith development in 50% of cases.³¹ Anemia can also develop due to the erosion of the malformation into the bowel lumen and subsequent bleeding, but also due to fragmentation of reticulocytes by the thrombus. The sequestration and coagulation can also lead to local or segmental bowel ischemia.¹⁶

Several reports describe malformations invading to surrounding structures, especially when the abnormality is in the rectosigmoid region with cases found involving the sacrum, bladder, and uterus. Despite the ability for local invasion, malignancy is rarely encountered.^{8,32,33}

CLINICAL PRESENTATION

History

Misdiagnosis is the theme with hemangiomas and malformations. Eighty percent of patients undergo one prior inappropriate surgical procedure.^{34–36} In a series with

Table 3 Syndromes that Are Associated with Hemangiomas and Vascular Malformations²⁰

Syndrome	Inheritance	Characteristics
Blue rubber bleb nevus syndrome	Most sporadic	Cavernous hemangiomas of the skin, GI tract, and other viscera. Lesions are blue, tender, and blanch.
Klippel-Trenaunay-Weber syndrome	Sporadic	Triad of cutaneous hemangiomas, bone, and soft tissue, hypertrophy of lower extremities, and congenital varicosities
Osler-Rendu-Weber syndrome	Autosomal dominant	Mucocutaneous telangiectasias, especially oral and nasal Hemangiomatous lesions in stomach, small intestine, and rectum

GI, gastrointestinal.

five patients, four had undergone a hemorrhoidectomy.³⁷ A series evaluating rectosigmoid cavernous hemangiomas had misdiagnosed the GI bleeding as hemorrhoids and ulcerative colitis.^{34,35} Another series of 47 patients had estimated a delay in diagnosis of 16 years.³⁸ Another review came to similar conclusions where the correct diagnosis was made at 19 years with 51% of patients undergoing an inappropriate and ineffective operation.³⁶

An estimated 80% of patients exhibit symptoms, with intraluminal bleeding in the majority of cases.^{39–41} Up to 90% have recurrent painless bleeding; half will have chronic iron-deficient anemia. The first presentation is often in childhood with bleeding episodes worsening with time.⁶ Although the majority of cases bleed into the lumen, there is potential for intraperitoneal or retroperitoneal bleeding when transmural malformations exist. Other non-GI malformations may also present with hemothorax or hemopericardium. Mucosal trauma causes erosion and eventually bleeding, with increased bleeding usually occurring in larger and distal subtypes.

Obstruction is also possible, though infrequent.⁴² Polypoid lesions act as a lead point for intussusception, or there may be luminal obstruction by circumferential masses.⁴³ Patients may experience constipation when tenesmus is seen with larger malformations. Abdominal or pelvic pain can be a common symptom.

Physical Examination

Hemangiomas and malformations provide few exam findings. Distal lesions can be detected on digital examination, though these masses are not usually overt. These tumors are soft and compressible, with a nodular sensation. When large, a mass on abdominal examination can sometimes be palpated. Table 3 details the syndromes associated with cutaneous manifestations.²⁰

Workup

LABORATORY

Evidence of chronic or acute blood loss will be present. In large consumptive masses, a decrease in clotting factors such as fibrinogen, platelets, and factors V and VIII is seen.

Imaging

PLAIN FILM

Sequestration can lead to calcified phleboliths that are evident in 50% of cases.³¹ The calcified phleboliths then can underestimate the presence of hemangiomas and malformations because the calcifications occur in the colonic wall, and do not extend into the soft tissue. The

Figure 2 Computed tomography reconstruction, simulating plain film imaging, shows the characteristic phleboliths and their typical distribution.

presence of these phleboliths, especially when lateral and outside the pelvic venous plexus, are specific signs of a venous malformation (Fig. 2).³³ Although rare in the normal population, phleboliths occur in less than 5% of individuals under 30; hence, if found in younger patients malformations may be a possible diagnosis.^{6,33,44}

Contrast Studies

Obstructing or polypoid lesions are the primary manifestation of hemangiomas and malformations identified by contrast studies (Fig. 3).³³ Anterior displacement of the rectum and widening of the presacral space can be

Figure 3 Barium enema. Multiple submucosal polypoid masses can be seen throughout, but especially in the rectosigmoid.

Figure 4 Computed tomography scan shows marked thickening of the rectosigmoid wall with phleboliths.

the result of the mass effect and soft tissue component of large cavernous rectal malformations. These masses may collapse with air insufflation.

Computed Tomography

On CT, pathognomonic findings consist of transmural enhancing bowel-wall thickening with or without phleboliths (Fig. 4).³³ The extent of extramural extension and surrounding invasion can also be accurately evaluated on a CT scan. One case review utilized CT colonography and concluded that it helped to identify mucosal lesions and intraluminal characteristics, as well as distribution.³³

Magnetic Resonance Imaging

MRI can also add to diagnosis, especially in situations of rectal malformations.⁸ On a T-2 weighted MRI, a high signal intensity can be seen, thought to be due to the slow flow. Increased signal intensity is also noted in the perirectal fat with serpiginous structures correlating to the small vessels supplying the hemangioma (Fig. 5). The findings mentioned are very specific findings, and not seen in any other clinical entities. That is why MRI can be more useful, perhaps, than CT. Although hemorrhoids can present with similar T2 findings, the location and lack of perirectal fat extension distinguishes hemorrhoids from rectal malformations. Phleboliths and calcifications are less easily detected on MRI versus CT or plain film.

ULTRASOUND

A vascular malformation presenting with massive hemorrhage during pregnancy was diagnosed with the use of endorectal ultrasound (Fig. 6). The diagnosis was aided by the utilization of Doppler studies demonstrating pulsatile flow.⁴⁵

Angiography

Mesenteric angiography has a characteristic pooling, most often seen in the rectosigmoid. This can be

Figure 5 T2-weighted magnetic resonance imaging shows rectal wall thickening (arrow) and perirectal serpiginous vascularity (star).

identified in the absence of active bleeding, and it is helpful for identifying synchronous lesions.^{6,7} A delayed venous phase is also a commonly seen pattern. However, the presence of thrombosis can lower the sensitivity of angiography as a diagnostic modality, as lesions can have a hypovascular or avascular appearance.

Endoscopy

Colonoscopy is crucial in the evaluation and workup of hemangiomas and malformations.^{40,46} As noted with air-contrast barium enemas, the polypoid lesions can collapse with insufflation. The intraluminal characteristics have submucosal projections that range from blue to red (Fig. 7). Pinpoint areas of bleeding are possible, with the presence of overt ulceration rarely seen. Mucosal edema, nodularity, and vascular congestion are present, and thus can be mistaken for the incorrect diagnosis of inflammatory bowel disease.^{2,17,31} Hemorrhoids are also a frequent misdiagnosis.

The upper GI tract should be evaluated to aid in the identification of synchronous lesions, and a complete colonoscopy done to assess the proximal extension. Biopsy is not recommended,^{6,17,18,31,44,47} due to the obvious potential for bleeding, although some have suggested biopsy with caution to promote an accurate diagnosis.⁴⁸

MANAGEMENT

Medical

As with all GI bleeding, critical care and resuscitative efforts take first priority to ensure that hemodynamic stability is achieved.

Figure 6 Endorectal ultrasound (ERUS) demonstrates sponge-like nature of malformation. Not well represented is the duplex flow that is specific for identifying the vascular component.

In true histologic hemangiomas, treatments with corticosteroids have been successful.^{49–51} However, most GI-associated hemangiomas are actually vascular malformations; therefore, pharmacological options are not effective.

Figure 7 Endoscopic view. A polypoid, purple nodule is seen. (Color not shown.)

Endoscopic

Ideal lesions of polypoid tumors with a narrow base have been successfully treated with snare polypectomy and cauterization.^{52–54} The argon beam coagulator has also been reported to have success, even in instances of severe hematochezia.⁵⁵ Over a course of 13 sessions, a moderate rectosigmoid malformation was successfully treated endoscopically with injections of *n*-butyl-2-cyanoacrylate.⁵⁶ The same authors also reported a case of similar lesion in a different patient, who required 15 sessions; the patient died from recurrent bleeding 4 months after treatment was completed. This technique should only be used if surgery is not a feasible option.

Operative

Despite more-conservative options, the treatment of choice is surgical resection. The recommended treatment for rectosigmoid malformations prior to 1971 was abdominoperineal resection.^{6,16,17,57–59} Because sphincter preservation is the goal, low anterior resection with mucosal resection is the current standard.⁶⁰ Other surgical options include segmental resection, a low anterior resection without a mucosectomy, or the modified Parks coloanal pull-through.^{61,62} Technically, the proximal margin is well

delineated by the presence of subserosal serpentine vessels in the colon with the malformation, as well as a more rigid bowel and thickened mesentery.⁶³ If normal distal bowel exists, a double-stapling approach is feasible.

In a mucosectomy, a sleeve may be resected based on the plane that exists between the muscularis and the mucosa.⁶⁴ Removal of the mucosa 0.5 cm proximal to the dentate line is made, with the aid of a submucosal epinephrine infiltration. Some approaches advocate a dissection down to the levators, the preservation of a 3 to 4 cm anal/distal cuff, and a hand-sewn anastomosis.⁴⁷ As with most anal anastomosis, a proximal diverting ileostomy is created.

In more proximal lesions, segmental resection, full-thickness wedge resection, suture ligation, and operative polypectomy are advised.⁹ The largest single institution analyzing blue-rubber bleb malformations advocates an aggressive surgical approach with eradication of all identified lesions. This is combined with interoperative push-enteroscopy to aid in a thorough evaluation of all malformations. Their series consisted of 10 patients, with rebleeding in one patient who had over 557 lesions. Their study advocates complete resection; lesions that were incompletely removed by banding or suture ligation techniques had expansion of residual malformations (rather than recurrence).

In an isolated proximal malformation, laparoscopic approaches have been successful.⁶⁵

CONCLUSION

Because they are uncommon and frequently misdiagnosed, an awareness of GI-associated hemangiomas and vascular malformations must be considered in situations of GI bleeding. Fortunately, diagnostic clues exist in the clinical, radiographic, and endoscopic workup to arrive at an etiology. Precision in the usage of the terms hemangiomas and vascular malformations should be emphasized to reflect the latest guidelines and classification. Surgery will continue to have a role, as minimally invasive techniques will help evolve the management.

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