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 entitities.[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 1:2.5[2]; 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 Malformations[11]

	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 I have used the term hemangioma 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[Q1].

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 Hemangioma Classification[15–19]

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

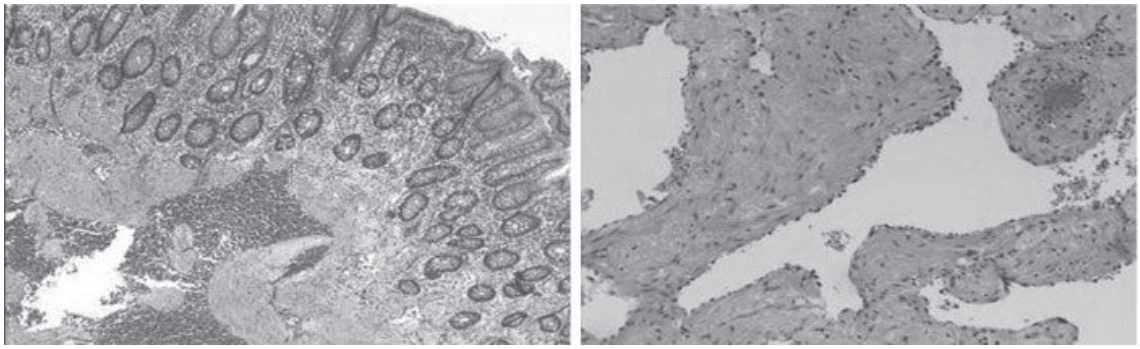


Figure 1 Cavernous vascular malformation. It is composed of blood-filled sinus-like spaces with prominent vascular channels in the submucosa. (Left: hematoxylin & eosin [H&E] 4x; right: H&E 10x).

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 hemangiomas developing from an earlier defect than cavernous hemangiomas in the stem cell cycle.[2,8,16]

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 hemangiomas 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 five patients, four had undergone a hemorrhoidectomy.[37]

Q2 **Table 3 Associated Syndromes[20]Q2**

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 blanche.
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.

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[Q3].[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 obscure the presence of hemangiomas and malformations because the calcifications occur in the colonic wall, and do not extend into the soft tissue. The presence of these phleboliths, especially when lateral and outside the pelvic venous plexus, are specific signs[Q4] (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]

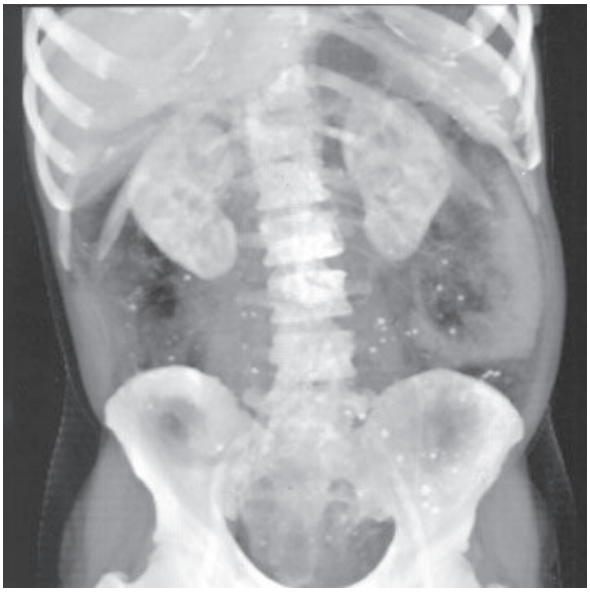


Figure 2 Computed tomography reconstruction, simulating plain film imaging, shows the characteristic phleboliths and their typical distribution.

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 the result of the mass effect and soft tissue component of

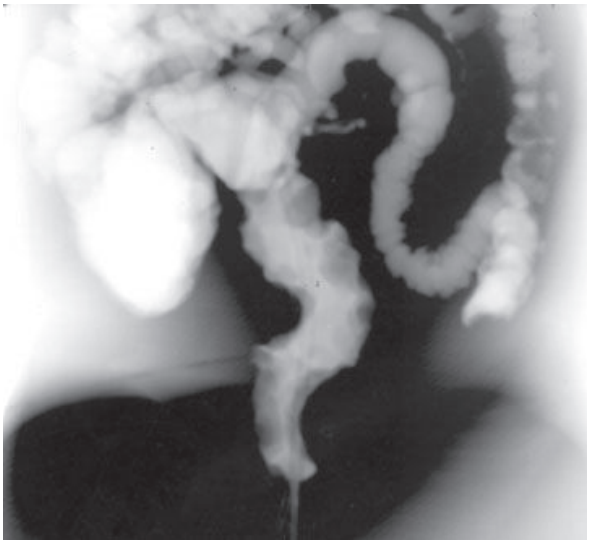


Figure 3 Barium enema. Multiple submucosal polypoid masses can be seen throughout, but especially in the rectosigmoid.

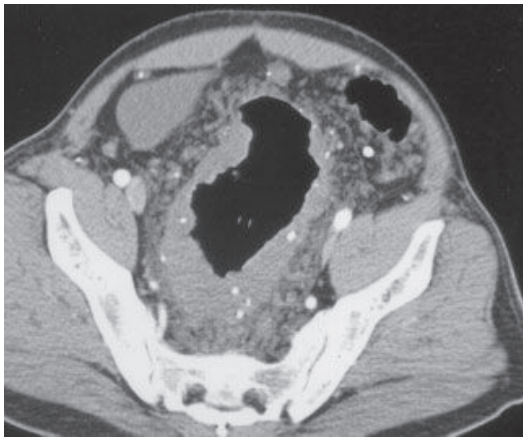


Figure 4 Computed tomography scan shows marked thickening of the rectosigmoid wall with phleboliths.

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 high T-2 weighted MRI, thickening[Q5] 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). MRI provides a higher specificity, especially over CT, which is particularly helpful in diagnosis. Although hemorrhoids can present with similar T2 findings, the location and lack of perirectal fat extension distinguishes hemorrhoids from rectal malformations[Q6]. Phleboliths and calcifications are less easily detected on MRI versus CT or plain film.

ULTRASOUND

A 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 identified in the absence of active bleeding, and it is

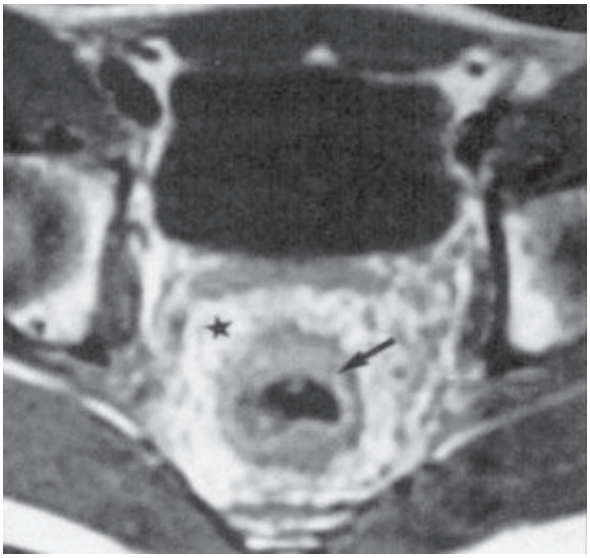


Figure 5 T2-weighted magnetic resonance imaging shows rectal wall thickening (arrow) and perirectal serpiginous vascularity (star).

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.

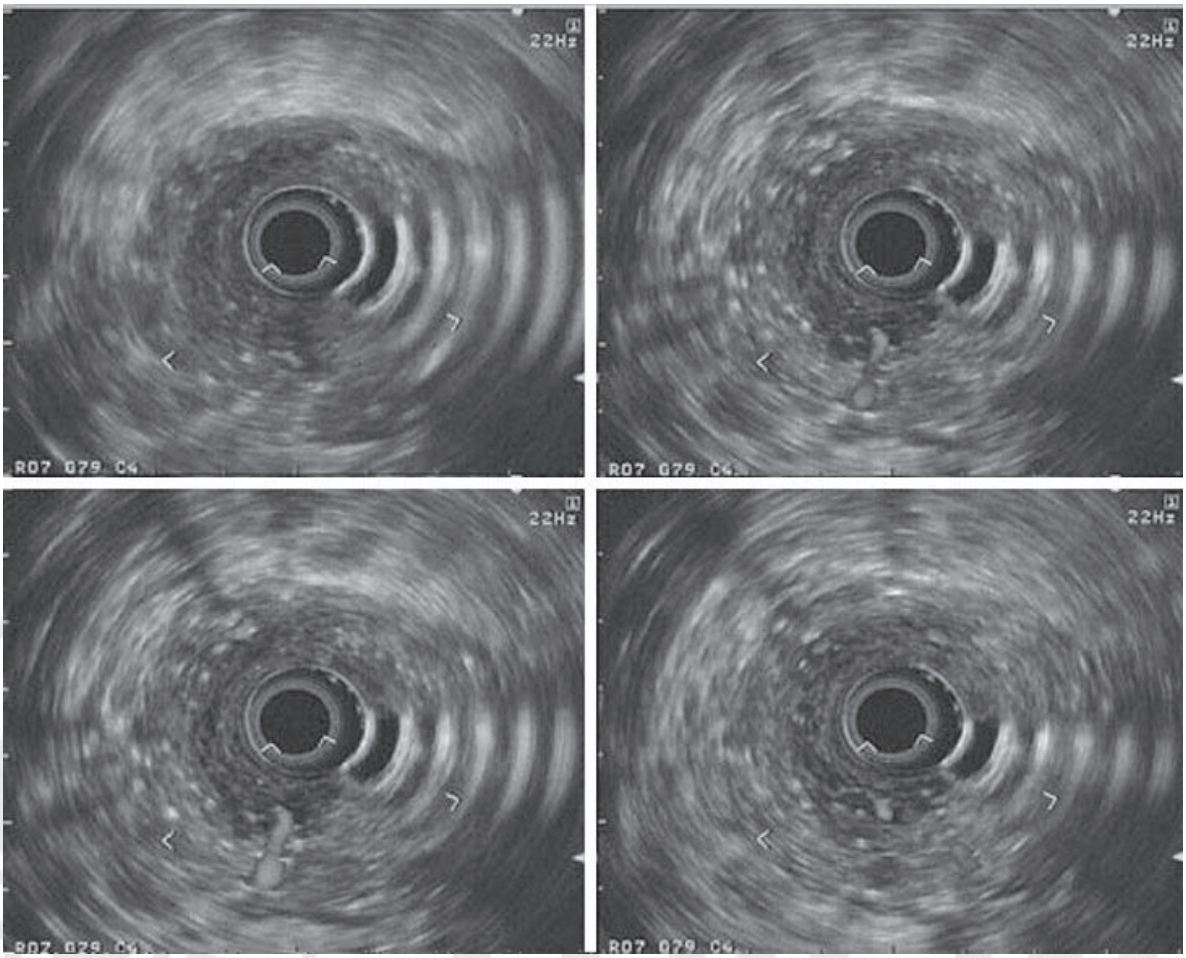
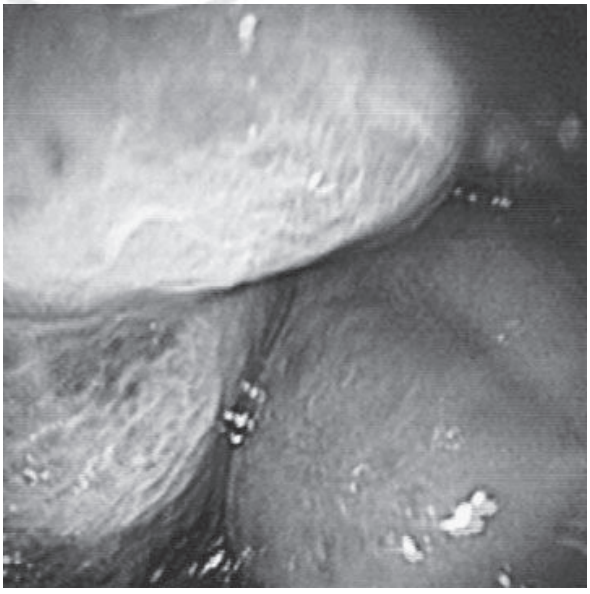


Figure 6 Endorectal ultrasound (ERUS) demonstrates sponge-like nature of malformation. Not well represented is the duplex flow that is specific for identifying the vascular component.



Q7 **Figure 7** Endoscopy[Q7].

In true histologic hemangiomas, treatment with corticosteroids has been successful.[49–51] However, most GI-associated hemangiomas are actually vascular malformations; therefore, pharmacological options are not effective.

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[Q8] 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.

Q8

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[Q9]. 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.[65Q10]

REFERENCES

1. Phillips B. Surgical cases. London Med Gaz 1839;23: 514–517
2. Gentry RW, Dockerty MB, Glagett OT. Vascular malformations and vascular tumors of the gastrointestinal tract. Surg Gynecol Obstet 1949;88(4):281–323
3. Varma JD, Hill MC, Harvey LAC. Hemangioma of the small intestine manifesting as gastrointestinal bleeding. Radiographics 1998;18(4):1029–1033
4. Lyon DT, Mantia AG. Large-bowel hemangiomas. Dis Colon Rectum 1984;27(6):404–414
5. Allred HW Jr, Spencer RJ. Hemangiomas of the colon, rectum, and anus. Mayo Clin Proc 1974;49(10):739–741[Q11]
6. Coppa GF, Eng K, Localio SA. Surgical management of diffuse cavernous hemangioma of the colon, rectum and anus. Surg Gynecol Obstet 1984;159(1):17–22
7. Dachman AH, Ros PR, Shekitka KM, Buck JL, Olmsted WW, Hinton CB. Colorectal hemangioma: radiologic findings. Radiology 1988;167(1):31–34
8. Djouhri H, Arrivé L, Bouras T, Martin B, Monnier-Cholley L, Tubiana JM. MR imaging of diffuse cavernous hemangioma of the rectosigmoid colon. AJR Am J Roentgenol 1998; 171(2):413–417
9. Fishman SJ, Smithers CJ, Folkman J, et al. Blue rubber bleb nevus syndrome: surgical eradication of gastrointestinal bleeding. Ann Surg 2005;241(3):523–528
10. Fernandez-Pineda I. Vascular tumors and malformations of the colon. World J Gastroenterol 2009;15(41):5242–5243
11. Mulliken JB, Glowacki J. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. Plast Reconstr Surg 1982; 69(3):412–422
12. Mallucci P. Vascular anomalies must be properly classified. BMJ 1999;319(7214):919
13. Mulliken J, Young AE. Vascular Birthmarks: Hemangiomas and Vascular Malformations. Philadelphia. WB Saunders; 1988:24–37
14. Marchuk DA. Pathogenesis of hemangioma. J Clin Invest 2001;107(6):665–666
15. Kaijser R. Uber hamangiome des tractus gastrointestinalis. Archiv Klin Chir 1936;187:351–388
16. Bland KI, Abney HT, MacGregor AMC, Hawkins IF. Hemangiomatosis of the colon and anorectum: case report and a review of the literature. Am Surg 1974;40(11):626–635
17. Chaimoff C. On the treatment of hemangioma of the rectum. Dis Colon Rectum 1985;28(8):632
18. Borum ML. Cavernous colorectal hemangioma: a rare cause of lower gastrointestinal bleeding and a review of the literature. Dig Dis Sci 1997;42(12):2468–2470
19. Pohlen U, Kroesen AJ, Berger G, Buhr HJ. Diagnostics and surgical treatment strategy for rectal cavernous hemangiomas based on three case examples. Int J Colorectal Dis 1999; 14(6):300–303
20. Tan MCB, Mutch MG. Hemangiomas of the pelvis. Clin Colon Rectal Surg 2006;19(2):94–101
21. Kempson RL, Fletcher CD, Evans HL, Hendrickson MR, Sibley RK. Atlas of Tumor Pathology, Third Series, Fascicle 30: Tumors of the Soft Tissues. Washington, DC: Armed Forces Institute of Pathology; 2001:307–367
22. Head HD, Baker JQ, Muir RW. Hemangioma of the colon. Am J Surg 1973;126(5):691–694
23. Pérez C, Andreu J, Llauger J, Valls J. Hemangioma of the rectum: CT appearance. Gastrointest Radiol 1987;12(4): 347–349
24. Rissier HL Jr. Hemangiomatosis of the intestine. Discussion, review of the literature and report of two new cases. Gastroenterologia 1960;93:357–385
25. Geschickter CF, Keasbey LE. Tumors of blood vessels. Am J Cancer 1935;23:568–591
26. Cheung DS, Warman ML, Mulliken JB. Hemangioma in twins. Ann Plast Surg 1997;38(3):269–274
27. Blei F, Walter J, Orlow SJ, Marchuk DA. Familial segregation of hemangiomatas and vascular malformations as an autosomal dominant trait: a rare genetic disorder. Arch Dermatol 1998;134:718–722[Q12]
28. Takahashi K, Mulliken JB, Kozakewich HP, Rogers RA, Folkman J, Ezekowitz RA. Cellular markers that distinguish the phases of hemangioma during infancy and childhood. J Clin Invest 1994;93(6):2357–2364[Q13]

29. Brizel HE, Raccuglia G. Giant hemangioma with thrombocytopenia. Radioisotopic demonstration of platelet sequestration. Blood 1965;26(6):751–764
30. Hall GW. Kasabach-Merritt syndrome: pathogenesis and management. Br J Haematol 2001;112(4):851–862
31. Aylward CA, Orangio GR, Lucas GW, Fazio VW. Diffuse cavernous hemangioma of the rectosigmoid—CT scan, a new diagnostic modality, and surgical management using sphincter-saving procedures. Report of three cases. Dis Colon Rectum 1988;31(10):797–802
32. Bortz JH. Diffuse cavernous hemangioma of the rectum and sigmoid. Abdom Imaging 1994;19(1):18–20
33. Hsu RM, Horton KM, Fishman EK. Diffuse cavernous hemangiomatosis of the colon: findings on three-dimensional CT colonography. AJR Am J Roentgenol 2002;179(4):1042–1044
34. Amarapurkar D, Jadliwala M, Punamiya S, Jhawer P, Chitale A, Amarapurkar A. Cavernous hemangiomas of the rectum: report of three cases. Am J Gastroenterol 1998;93(8):1357–1359
35. Levy AD, Abbott RM, Rohrmann CA Jr, Frazier AA, Kende A. Gastrointestinal hemangiomas: imaging findings with pathologic correlation in pediatric and adult patients. AJR Am J Roentgenol 2001;177(5):1073–1081
36. Oner Z, Altaca G. Diffuse cavernous rectal hemangioma—clinical appearance, diagnostic modalities and sphincter saving approach to therapy: report of 2 and a collective review of 79 cases. Acta Chir Belg 1993;93(4):173–176
37. Wang HT, Tu Y, Fu CG, et al. Diffuse cavernous hemangioma of the rectosigmoid colon. Tech Coloproctol 2005;9(2):145–148
38. Nader PR, Margolin F. Hemangioma causing gastrointestinal bleeding. Case report and review of the literature. Am J Dis Child 1966;111(2):215–222
39. Sylla P, Deutsch G, Luo J, et al. Cavernous, arteriovenous, and mixed hemangioma-lymphangioma of the rectosigmoid: rare causes of rectal bleeding—case series and review of the literature. Int J Colorectal Dis 2008;23(7):653–658
40. Fenoglio-Preiser CM, Pascal RR, Perzin KH. Atlas of Tumor Pathology, Second Series, Fascicle 27: Tumors of the Intestines. Washington, DC: Armed Forces Institute of Pathology; 1990:473–483
41. Babcock WW, Jonas KC. Hemangioma of the colon. Am J Surg 1950;80(7):854–859
42. Buie LA, Swan T. Benign tumors of the colon. Surg Clin North Am 1929;9:893–910
43. Sawyer CF. Hemangioma of colon. Arch Surg 1939;39: 987–991
44. Stening SG, Heptinstall DP. Diffuse cavernous haemangioma of the rectum and sigmoid colon. Br J Surg 1970; 57(3):186–189
45. Gottlieb K, Coff P, Preiksaitis H, Juviler A, Fern P. Massive hemorrhage in pregnancy caused by a diffuse cavernous hemangioma of the rectum—EUS as imaging modality of choice. Medscape J Med 2008;10(9):206
46. Yorozuya K, Watanabe M, Hasegawa H, et al. Diffuse cavernous hemangioma of the rectum: report of a case. Surg Today 2003;33(4):309–311
47. Cunningham JA, Garcia VF, Quispe G. Diffuse cavernous rectal hemangioma—sphincter-sparing approach to therapy. Report of a case. Dis Colon Rectum 1989;32(4):344–347
48. Wang CH. Sphincter-saving procedure for treatment of diffuse cavernous hemangioma of the rectum and sigmoid colon. Dis Colon Rectum 1985;28(8):604–607
49. Bartoshesky LE, Bull M, Feingold M. Corticosteroid treatment of cutaneous hemangiomas: how effective? A report on 24 children. Clin Pediatr (Phila) 1978;17(8):625–638, 629–638
50. Akyüz C, Yari N, Kutluk MT, Büyükpamukçu M. Management of cutaneous hemangiomas: a retrospective analysis of 1109 cases and comparison of conventional dose prednisolone with high-dose methylprednisolone therapy. Pediatr Hematol Oncol 2001;18(1):47–55
51. Ezekowitz RAB, Mulliken JB, Folkman J. Interferon alfa-2a therapy for life-threatening hemangiomas of infancy. N Engl J Med 1992;326(22):1456–1463
52. Fraiberg EN, Ahmed S. Colonoscopic excision of a polypoidal cavernous hemangioma of the cecum. Gastrointest Endosc 1985;31(2):109
53. Liang LC, Forbes N, David J, Ozick L. Endoscopic polypectomy of an unusually long polypoid colorectal cavernous hemangioma. Gastrointest Endosc 1998;47(3): 307–308
54. Levitt RE. Colonic cavernous hemangioma. Gastrointest Endosc 1998;48(3):337
55. Benson JM, Orlay G. Colorectal haemangioma and its relationship to haemorrhoids in childhood. Aust N Z J Surg 1991;61(7):537–540
56. Zurakowski J, Swiercz P, Wróblewski T, et al. Diffuse cavernous hemangioma of rectosigmoid colon treated with n-butyl-2-cyanoacrylate injections. Endoscopy 2008;40 (Suppl 2):E120–E121
57. Jaques AA. Cavernous hemangioma of the rectum and rectosigmoid colon. Am J Surg 1952;84(5):507–509
58. Hellstrom J, Hultborn KA, Engstedt L. Diffuse cavernous hemangioma of the rectum. Acta Chir Scand 1955;109(3-4): 277–283
59. Westerholm P. A case of diffuse haemagiomatosis of the colon and rectum. Acta Chir Scand 1967;133:173–176
60. Demircan O, Sönmez H, Zeren S, Coar E, Bicakci K, Ozkan S. Diffuse cavernous hemangioma of the rectum and sigmoid colon. Dig Surg 1998;15(6):713–715
61. Jeffery PJ, Hawley PR, Parks AG. Colo-anal sleeve anastomosis in the treatment of diffuse cavernous haemangioma involving the rectum. Br J Surg 1976;63(9): 678–682
62. Parks AG. Benign tumors of the rectum. In: Rob C, Smith R, Morgan CN eds. Clinical Surgery. Vol 10. Abdomen and Rectum and Anus. London: Butterworths; 1966:541–548
63. Londono-Schimmer EE, Ritchie JK, Hawley PR. Coloanal sleeve anastomosis in the treatment of diffuse cavernous haemangioma of the rectum: long-term results. Br J Surg 1994;81(8):1235–1237
64. Telander RL, Ahlquist D, Blaufuss MC. Rectal mucosectomy: a definitive approach to extensive hemangiomas of the rectum. J Pediatr Surg 1993;28(3):379–381
65. Huh JW, Cho SH, Lee JH, Kim HR. Large cavernous hemangioma in the cecum treated by laparoscopic ileocecal resection. World J Gastroenterol 2009;15(26):3319–3321