Turk J Gastroenterol 2008; 19 (4): 271-275

Rapidly growing giant solitary cavernous hemangioma in a patient with ulcerative colitis

Ülseratif kolitli bir hastada hızlı büyüyen soliter dev kavernöz hemanjiom

Gülbanu ERKAN[1], Hakan ALAGÖZLÜ[1], Leyla MEMİŞ[2], Candan TUNCER[1]

Departments of [1]Gastroenterology and [2]Pathology, Gazi University, School of Medicine, Ankara

Cavernous hemangiomas are rare lesions of the colon that usually present with painless, recurrent bleeding. Hemangiomas can be capillary or cavernous type, and 80% of rectal hemangiomas are cavernous type. Ulcerative colitis is an inflammatory disorder that affects the rectum and, occasionally, the whole colon. Diarrhea, rectal bleeding and mucous discharge characterize ulcerative colitis. We present a 61-year-old man with painless rectal bleeding due to a solitary cavernous hemangioma of the rectum. He had been diagnosed with distal type ulcerative colitis in 2003. He was asymptomatic under mesalazine treatment until May 2005, when he presented with new onset bright red rectal bleeding and mucous discharge, despite still defecating normal stools once a day. Rectosigmoidoscopic examination revealed mucosal hyperemia, edema, granularity, and a hyperemic, friable mass lesion 5x4 cm in diameter in the rectum. Following excision, histopathologic examination of the mass was consistent with cavernous hemangioma. There was a six-month period between the rectosigmoidoscopy in which the cavernous hemangioma (5x4 cm in diameter) was detected and the former rectosigmoidoscopy with no reported hemangioma. Thus, this was considered a rapidly growing cavernous hemangioma. Intralesional microhemorrhages may cause rapid enlargement of the hemangiomas. Ulcerative colitis is characterized by inflammation, which may interfere with vascular integrity and augment intralesional microhemorrhage. We postulate that the inflammatory background of ulcerative colitis may have accelerated intralesional hemorrhage and growth of this coincidental rectal cavernous hemangioma. To the best of our knowledge, this is the only case of this sort in the literature.

Key words: Ulcerative colitis, cavernous hemangioma, rectal bleeding

Kavernöz hemanjiomlar kolonun nadir görülen lezyonları olup, genellikle ağrısız, rekürren kanama ile prezante olurlar. Hemanjiomlar kapiller ya da kavernöz tip olabilirler; rektal hemanjiomların % 80'i kavernöz tiptir. Ülseratif kolit rektumu ve bazen de bütün kolonu tutan inflamatuar bir hastalıktır. Diyare, rektal kanama ve mukuslu dışkılama ülseratif kolitin başlıca özellikleridir. Olgumuz, rektumdaki soliter kavernöz hemanjioma bağlı ağrısız rektal kanaması olan 61 yaşında bir erkek hastadır. Hastaya 2003 yılında distal tip ülseratif kolit tanısı konulmuştu. Hasta, yeni başlangıçlı parlak kırmızı renkli rektal kanama ve mukus deşarjı şikayetleriyle başvurduğu 2005 yılı Mayıs ayına kadar mesalazine tedavisi ile asemptomatikti. Bu yakınmaların başlamasına rağmen, hasta halen günde bir defa, normal vasıfta defekasyon tanımlıyordu. Rektosigmoidoskopik incelemede mukozal hiperemi, ödem, granülarite ve rektumda 5 x 4 cm boyutlarında, hiperemik, frajil bir kitle lezyonu saptandı. Kitlenin cerrahi eksizyonu sonrasında yapılan histopatolojik inceleme kavernöz hemanjiom ile uyumluydu. 5 x 4 cm boyutlarında bir kavernöz hemanjiomun tespit edildiği rektoskopi ile, hemanjiom tespit edilmeyen bir önceki rektoskopi arasındaki süre 6 aydır. Dolayısıyla, kavernöz hemanjiomun hızlı büyüdüğü kanısına varılmıştır. İntralezyonel mikrohemoraji, kavernöz hemanjiomların hızla büyümesine yol açabilir. Ülseratif kolit inflamasyonla karakterize olduğundan, vasküler bütünlüğü bozarak intralezyonel mikrohemorajiyi arttırabilir. Bizim hipotezimiz, ülseratif kolite bağlı inflamasyon zemininin intralezyonel mikrohemorajiyi ve bu ko-insidental rektal kavernöz hemanjiomun büyümesini hızlandırmış olabileceği şeklindedir. Bildiğimiz kadarıyla, bu tür bir olgu literatürde ilk defa bildirilmektedir.

Anahtar kelimeler: Ülseratif kolit, kavernöz hemanjiom, rektal kanama

INTRODUCTION

Colonic cavernous hemangiomas, which are most frequently localized in the rectum and the sigmoid colon, are encountered rarely. The most common symptom is painless, recurring rectal bleeding. If the hemangioma reaches a critical size, it can cause diarrhea, abdominal cramps and intestinal obstruction.

Hemangiomas can be capillary or cavernous type, and 80% of rectal hemangiomas are cavernous type. They can be solitary or multiple, and they

Address for correspondence: Gülbanu ERKAN
Gazi Üniversitesi Tıp Fakültesi
Gastroenteroloji Bilim Dalı Ankara, Turkey
Phone: + 90 312 202 58 22 • Fax: + 90 312 223 63 65
E-mail: gcanbaloglu@yahoo.com

Manuscript received: 12.07.2007 **Accepted:** 13.03.2008

may also cause diffuse involvement of the gastrointestinal tract. Diagnostic tests for cavernous hemangiomas are endoscopic examination and angiography.

Ulcerative colitis is an inflammatory disorder that affects the rectum, and the entire colon may also be involved. The most common symptoms are diarrhea, rectal bleeding and mucous discharge.

Lesion growth is accelerated by intralesional hemorrhage in cavernous hemangiomas. The mucosal inflammation of ulcerative colitis may augment intralesional hemorrhage and growth of a coincidental cavernous hemangioma.

CASE REPORT

A 61-year-old man was admitted in our clinic with bright red rectal bleeding and mucous stools. He had presented in 2003 with mucous diarrhea containing bright red blood once or twice a day. The colonoscopy had revealed mucosal hyperemia, edema, friability, and granularity in the rectum and sigmoid colon, which were interpreted as distal type ulcerative colitis. Biopsy findings were consistent with the endoscopic diagnosis. His rectal bleeding and mucous discharge had responded well to mesalazine enema, which he had been using for two years. His medical history included systemic arterial hypertension and hyperlipidemia, which were controlled with dietary measures, and he had also undergone a hemorrhoidectomy 20 years ago. He was asymptomatic with once daily mesalazine enema treatment until May 2005, when he presented with new onset bright red colored rectal bleeding and mucous discharge. He was still defecating normal stools once a day, and rectal bleeding was small in amount and dripping. His physical examination was normal. His complete blood count was normal, and his blood biochemistry was normal except for a fasting blood glucose of 140 mg/dl and a total cholesterol of 225 mg/dl. His erythrocyte sedimentation rate was 37 mm/hour and C-reactive protein was 43 mg/L (normal range in our laboratory: 0-6 mg/L). His stool examination was negative for parasites, amoeba, and Giardia. Wright staining revealed 1-2 leukocytes in each field. Rectosigmoidoscopic examination revealed mucosal hyperemia, edema, granularity, scarce erosions, and a hyperemic, friable mass lesion 5x4 cm in diameter in the rectum (Figure 1). Histopathologic examination of the biopsy material obtained from the mass lesion was consistent with prominent crypt distortion characterized by atrophy, mononuclear inflammatory cell infiltration in the lamina propria, focal cryptitis with polymorphonuclear neutrophils and eosinophils, and pronounced regenerative activity. Although no dysplastic changes were observed, close follow-up with frequent biopsies was recommended because of scarce p53 expression in the cryptic endothelial cells. Histopathologic findings were interpreted as ulcerative colitis showing signs of activity. Although the histopathologic examination of the mass lesion did not reveal any manifest malignancy, rapid growth of the mass and scarce p53 expression raised suspicion of a malignant tumor. The mass was surgically excised, and was 5x4x2.5 cm and brownish black in color. The cross-section of the mass consisted of vascular structures of various sizes containing hemorrhage. Histopathologic findings were consistent with cavernous hemangioma (Figure 2).

Rectal bleeding and mucous discharge disappeared after the excision. In the follow-up period, repeat rectosigmoidoscopy revealed mild hyperemia, edema, granularity, friability with the touch of the endoscope, and internal hemorrhoids. Histopathology revealed prominent atrophy in the crypts,

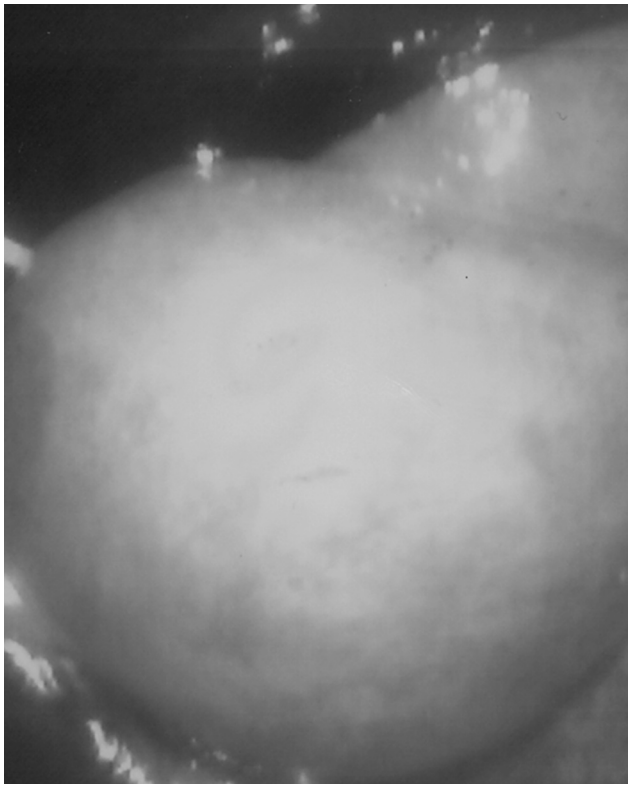

Figure 1. Rectosigmoidoscopic image of the cavernous hemangioma.

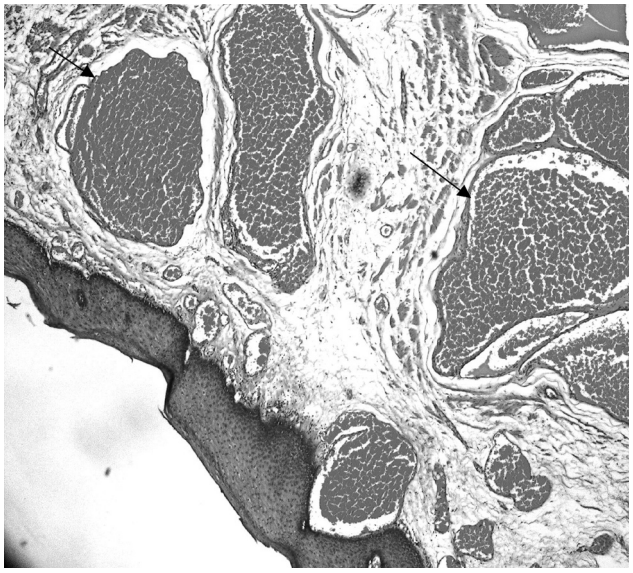

Figure 2. Cavernous hemangioma formed by congested and dilated vascular structures (hematoxylin & eosin, x 40).

crypt distortion, and mononuclear inflammatory cell infiltration in the lamina propria with rare polymorphonuclear leukocytes; findings were consistent with ulcerative colitis in the inactive phase. The patient is currently under follow-up, and is asymptomatic 18 months after surgery.

DISCUSSION

Vascular lesions of the colon are classified as benign or malignant lesions. Benign lesions are capillary and cavernous hemangiomas. Colonic cavernous hemangiomas are rare lesions, most frequently localized in the rectum and the sigmoid colon (2). They were first described by Phillips in 1839 (3). The most common symptom is painless, recurring rectal bleeding. If the lesion reaches a critical size it can cause diarrhea, abdominal cramps and intestinal obstruction (4,5). Cavernous hemangiomas of the colon present with rectal bleeding in more than 75% of the cases. Bleeding may be occult or massive; it is episodic and frequently occurs in the early stages of life. They can mimic internal hemorrhoids, ulcerative colitis, and cancer because of intermittent rectal bleeding (6).

Hemangiomas are rare lesions of the colon (6). Diagnostic tests for rectal hemangioma are endoscopic examination, in which biopsy material is obtained for histopathologic examination, and angiography. Biopsy is related to risk of bleeding, which may rarely necessitate emergency surgery. It has been reported that 28.6% of patients with known or suspected cavernous hemangiomas develop massive bleeding (7). Phleboliths and displacement or distortion of the rectal air column are characteristic plain X-ray findings. In endoscopic examination, red nodules elevated from the mucosal surface and vascular congestion are observed, and ulcerations and proctitis may also be seen. Angiography confirms the diagnosis of hemangioma, and visceral angiography is useful for ruling out coincidental hemangiomas elsewhere in the gastrointestinal tract.

Hemangiomas can be capillary or cavernous; 80% of rectal hemangiomas are of cavernous type. Hemangiomas can be solitary or multiple, even the whole gastrointestinal tract may be diffusely involved. Differential diagnosis includes internal hemorrhoids, inflammatory bowel disease, carcinoma, and various other pathological conditions (5,6,8). Jeffery et al. (9) reported a series of patients, 80% of whom had undergone at least one inappropriate surgical procedure because of incorrect diagnosis until the diagnosis of rectal hemangioma was established. Hence, choosing the appropriate tests for the diagnosis of cavernous hemangioma is of utmost importance. The characteristic appearances in the barium X-ray are well defined but non-specific (10). Mesenteric angiography is a useful but invasive method and the lesion may appear hypovascular or hypervascular depending on whether or not the lumen of the hemangioma is thrombosed (11). Radiologically visualized phleboliths are present in 50% of adults, rarely in children, and are an important clue in diagnosis (12). Endoscopy is the method of choice for diagnosis (13). Although proctosigmoidoscopy may be adequate for establishing the diagnosis in some cases, a definite diagnosis usually requires a biopsy despite the risk of hemorrhage. Computed tomography is a non-invasive technique that may visualize phleboliths that a plain X-ray can miss (14). In addition, computed tomography gives important information about the dimension of longitudinal bowel involvement, the degree of bowel wall thickening, involvement of the adjacent pelvic viscera, and the number and vascularity of lesions (15). Magnetic resonance imaging (MRI) can also be used for diagnosis; rectal surface coil MRI gives more detailed information regarding the five layers of the bowel wall and anal sphincter muscles when compared to conventional MRI (16).

Rectal hemangiomas are rare lesions and they are often misdiagnosed because they may mimic inf-

lammatory bowel disease, internal hemorrhoids, carcinoma, and various other pathological conditions.

There was a six-month period between the rectosigmoidoscopy in which we detected the cavernous hemangioma and the former rectosigmoidoscopy with no reported hemangioma. Therefore, it was decided that the cavernous hemangioma was a rapidly growing one, with a diameter of about 5x4 cm. To the best of our knowledge, such rapid growth and concurrence with ulcerative colitis has not been previously reported for a cavernous hemangioma. Surgical excision of the cavernous hemangioma resulted in remission of bleeding and mucous discharge, and the patient is currently asymptomatic with mesalazine enema treatment for his ulcerative colitis. The recurrence of bleeding and mucous discharge in a patient who is in the remission phase of ulcerative colitis must bring to mind the possibility of concurrent cavernous hemangioma. In this setting, proctosigmoidoscopy is often necessary for discrimination of activation of ulcerative colitis from a coexisting rectal cavernous hemangioma. Biopsy may be required to discriminate the cavernous hemangioma from carcinoma and other polypoid lesions.

Our patient's hemangioma reached a diameter of 5x4 cm over six months. This rapid growth was misleading because it raised suspicions of an aggressive tumor, before the histopathology revealed a cavernous hemangioma. The growth of cavernous hemangiomas has been attributed to the higher tendency of intracranial cavernous hemangiomas to undergo repeated intralesional microhemorrhage. The breakdown products of microhemorrhage may promote formation of leaky vessels, which leads to further hemorrhage and a vicious cycle of growth (17, 18). Ulcerative colitis may contribute to repeated intralesional microhemorrhage because of colonic mucosal inflammation. Also, endothelium of the hemangioma shares molecular phenotype with blood cells. LMO2 is a transcriptional regulator that plays an important role in angiogenesis. It is logical to postulate that LMO2 may play a role in the pathogenesis and proliferation of hemangiomas. LMO2-associated protein complexes could regulate angiogenesis (19). The endothelia of hemangiomas may express various markers to escape the immune surveillance. Recent research reported that CD8+ T cells in hemangiomas and the endothelia of hemangioma uniquely expressed leukocyte marker Fcgamma receptor IIb (FCgammaRII) (20). An immune response may be one of the mechanisms for hemangioma proliferation in concurrence with the ulcerative colitis.

In conclusion, we hypothesize that mucosal inflammation associated with ulcerative colitis may have contributed to intra-lesional microhemorrhage and the unusually rapid growth of the coincidental rectal cavernous hemangioma in this patient. Cavernous hemangioma is a rare entity, and it should be considered in the differential diagnosis of rectal bleeding in patients with ulcerative colitis who are in the remission phase.

REFERENCES

1. Amarapurkar D, Jadliwala M, Punamiya S, et al. Cavernous hemangiomas of the rectum: report of three cases. Am J Gastroenterol 1998;93:1357–9.
2. Sweeney K, Petrelli N, Herrera L, et al. Cavernous hemangioma of the anus. J Surg Oncol 1984;27:286–8.
3. Demirbas T, Guler N, Caliskan C, et al. Mechanical bowel obstruction due to colonic hemangioma: report of a case. Turk J Gastroenterol 2006;17:305-7.
4. Gentry RW, Dockerty MB, Clagett OT. Vascular malformations and vascular tumours of the gastrointestinal tract. Int Abstr Surg 1949;88:281–323.
5. Allred HW, Spencer RJ. Hemangiomas of the colon, rectum and anus. Mayo Clin Proc 1974;49:739–41.
6. Stening SG, Heptinstall DP. Diffuse cavernous hemangioma of the rectum and sigmoid colon. Br J Surg 1970;57: 186-9.
7. Oner Z, Altaca G. Diffuse cavernous rectal hemangioma - clinical appearance, diagnostic modalities and sphincter saving approach to therapy: report of 2 and collective review of 79 cases. Acta Chir Belg 1993;93:173–6.
8. Lyon DT, Mantia AG. Large bowel hemangioma. Dis Colon Rectum 1984;27:404–14.
9. Jeffery PJ, Hawley PR, Parks AG. Colo-anal sleeve anastomosis in the treatment of diffuse cavernous hemangioma involving the rectum. Br J Surg 1976;63:678–82.
10. Bell GA, McKenzie AD, Emmons H. Diffuse cavernous hemangioma of the rectum: report of a case and review of the literature. Dis Colon Rectum 1972;15:377-82.
11. Dachman AH, Ros PR, Shekitka KM, et al. Colorectal hemangioma: radiologic findings. Radiology 1988;167:31–4.
12. Bland KI, Abney HT, MacGregor AMC, Hawkins IF. Hemangiomatosis of the colon and anorectum. Am Surg 1974; 40:626–35.
13. Bugra D, Bulut T, Yamaner S, et al. Soft-tissue images. Diffuse cavernous rectal hemangioma. Can J Surg 2002;45:365-6.
14. Bortz JH. Diffuse cavernous hemangioma of the rectum and sigmoid. Abdom Imaging 1994;19:18–20.
15. Sharma S, Gandhi D, Thulkar S, Dwivedi DN. Role of computed tomography in diagnosis of diffuse cavernous hemangioma of the rectum. Am J Gastroenterol 1999;94:1986–7.

16. Yorozuya K, Watanabe M, Hasegawa H, et al. Diffuse cavernous hemangioma of the rectum: report of a case. Surg Today 2003;33:309–11.
17. Pozzati E, Acciarri N, Tognetti F, et al. Growth, subsequent bleeding, and de novo appearance of cerebral cavernous angiomas. Neurosurgery 1996;38:662–9.
18. Rivera PP, Willinsky RA, Porter PJ. Intracranial cavernous malformations. Neuroimaging Clin N Am 2003;13:27–40.
19. Sun ZJ, Zhang L, Zhao YF. A postulated role for transcriptional regulator LMO2 in the proliferation and involution of hemangioma. Med Hypotheses 2006;67:1230-2.
20. Sun ZJ, Zhao YF, Zhang WF. Immune response: a possible role in the pathophysiology of hemangioma. Med Hypotheses 2007;68:353-5.