

Giant Cavernous Hemangioma of Small Intestine Mesentery: A Rare Cause of Recurrent Acute Symptomatic Anemia

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Gastrointestinal hemangiomas are rare vascular hamartomatous neoplasms. The cavernous type is the most common. Few cases of giant cavernous hemangiomas of small intestine mesentery have been reported. It is unclear whether they originate from the mesentery and extend into the intestinal wall or vice versa.

We present an adult male who was readmitted to several medical centers due to recurrent acute symptomatic anemia, without overt gastrointestinal hemorrhage. Thorough investigation was inconclusive. Surgery was both diagnostic and therapeutic. Giant cavernous hemangioma of the small intestinal mesentery was concluded. There was no evidence of recurrence after 1 year.

PATIENT DESCRIPTION

A 53-year-old male presented to our surgical outpatient clinic following readmission to several medical centers due to recurrent complaints of sudden weakness, dizziness, and muscle cramps. The first episode occurred 15 years ago, and four recurrent episodes occurred during the past 16 months. During each incident, the physical examination was normal without overt gastrointestinal hemorrhage, and hemoglobin levels dropped up to 5 g/dl,

requiring blood transfusions. The patient was asymptomatic between episodes, with mild chronic anemia.

Investigation was thorough yet discontinuous. Upper gastrointestinal endoscopy revealed erosive gastritis, and *Helicobacter pylori* was eradicated. Lower gastrointestinal endoscopy was normal. Video capsule endoscopy showed fresh blood 104 minutes after capsule ingestion (i.e., after 50 minutes in the small intestine); however, the bleeding source could not be determined. Computed tomography depicted a cluster of masses in the small intestine mesentery [Figure 1A]. Two percutaneous biopsies were inconclusive. Bone marrow biopsy demonstrated active trilineage hematopoiesis.

Following a prolonged course of investigation and readmissions, the patient presented to our surgical outpatient clinic. Physical examination revealed a right lower quadrant mass, and diagnostic laparoscopy was scheduled. The patient experienced another symptomatic episode prior to surgery, with hemoglobin levels dropping to 6.1 g/dl from 11 g/dl 3 weeks earlier. He received blood transfusions and underwent surgery. Laparoscopy revealed a 25 cm bluish-purple vascular neoplasm in the mesentery of the mid small intestine [Figure 1B]. Surgery was converted to laparotomy, and segmental small bowel resection with complete mesenteric excision was performed. The postoperative course was uneventful.

Macroscopic examination of the specimen revealed a sponge-like blood-filled vascular neoplasm located mostly in the mesentery of the small intestine [Figure 1C],

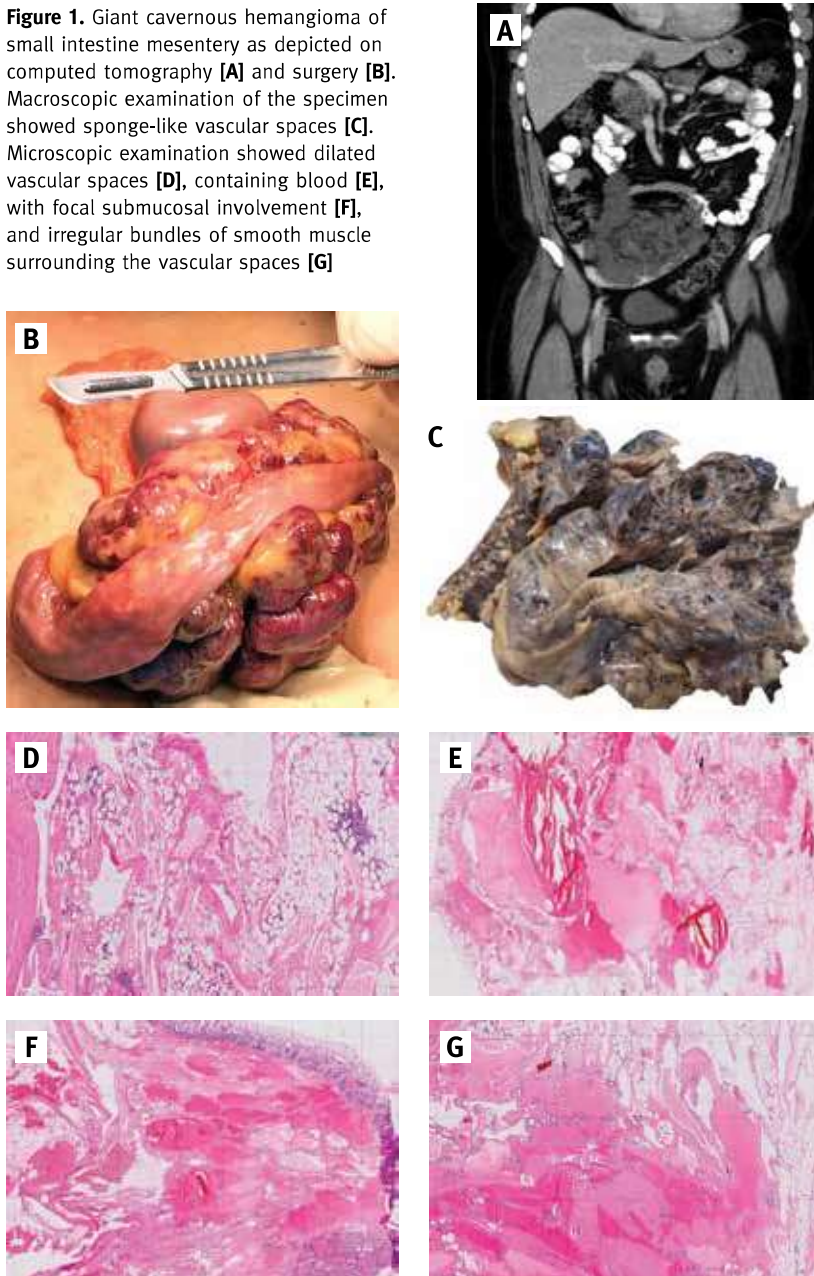
with focal involvement of the intestinal wall. The mucosa was intact. On microscopic examination [Figure 1D–1G] the neoplasm was shown to be composed of dilated thin-walled blood-filled vascular spaces, typical of cavernous hemangiomas. These spaces were lined by endothelial cells without atypia or mitotic activity. In some areas, irregular bundles of smooth muscle actin-positive cells were seen surrounding the vascular spaces, characteristic of venous hemangiomas. The submucosa of the small intestine was focally involved. On immunohistochemistry, the endothelial cell lining stained positive for vascular endothelial markers CD31 and ERG, whereas staining was negative for D2-40, HHV-8, HMB-45, and Melan A. There was no over-expression of MDM-2. The diagnosis was giant cavernous hemangioma of small intestine mesentery mixed with few areas of venous hemangioma.

One year after surgery the patient was found to be asymptomatic, had hemoglobin level of 16.5 g/dl, and was disease free, as determined during laparoscopic postoperative ventral hernia repair.

COMMENT

Hemangiomas are rare vascular hamartomatous neoplasms. They originate from embryonic sequestrations of mesodermal tissue and appear in ectopic locations [1,2]. Gastrointestinal hemangiomas are rare. They are classified into capillary, cavernous, and mixed types. Cavernous gastrointestinal hemangiomas are the most common type, and arise from larger submucosal vascular plexuses.

Figure 1. Giant cavernous hemangioma of small intestine mesentery as depicted on computed tomography [A] and surgery [B]. Macroscopic examination of the specimen showed sponge-like vascular spaces [C]. Microscopic examination showed dilated vascular spaces [D], containing blood [E], with focal submucosal involvement [F], and irregular bundles of smooth muscle surrounding the vascular spaces [G]



Giant cavernous hemangiomas of small intestine mesentery are extremely rare. Most reports involved both the mesentery and intestine, whereas the current case showed mostly in the mesentery, as described by Yang et al. [3]. It is unclear whether they originate from the mesentery and extend into the intestine or vice versa.

These bluish-purple vascular neoplasms consist of large blood-filled thin-walled sinuses, which are lined with endothelial cells and separated by minimal connective tissue [2]. The current case showed few areas of less organized smooth muscle bundles, characteristic of venous hemangiomas [1,3].

Cavernous hemangiomas might present as overt gastrointestinal hemorrhage (hematochezia or melena), chronic anemia, abdominal pain, palpable mass, or hemo-peritoneum. To the best of our knowledge, ours is the first report of giant cavernous mesenteric hemangioma manifesting with recurrent acute severe symptomatic anemia. Negative gastrointestinal endoscopy suggests further investigation with video capsule endoscopy and intravenous contrast enhanced computed tomography [4,5]. Exploratory laparoscopy should be considered as an integral part of diagnostic measures. Segmental small bowel resection with complete mesenteric excision is the treatment of choice.

CONCLUSIONS

The current case report underscores the crucial role of surgery as a diagnostic means for cavernous hemangiomas of small intestine mesentery and also for definitive treatment. This extremely rare benign neoplasm should be considered in the differential diagnosis of acute anemia.

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