Henry Ford Hospital Medical Journal

Volume 19 | Number 4 Article 4

12-1961

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Henry Ford Hosp. Med. Journal
Vol. 19, No. 4, 1971

Massive Gastrointestinal Hemorrhage Originating from the Cecum *

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Six cases are reported of arterio-venous malformations of the cecum presenting with hemorrhage. Newer techniques of diagnosis and management are reviewed. These cases are of interest because of their relative uniqueness and clinical importance. The diagnosis of underlying disease in these cases is often difficult. Massive bleeding, if not controlled, is an indication for immediate operation. Recurrent bleeding, even if previously controlled, can also be considered a surgical indication.

The cecum becomes a surgical problem most commonly because of clinical or radiological manifestations produced by a neoplastic lesion. Most of the symptoms are chronic. Obstruction is rare or late with right-sided lesions, but common in the left colon. Consideration must be given also to less common non-neoplastic lesions when symptoms are present of a specific inflammatory nature such as tuberculosis, polyps, granulomatous colitis and cecal involvement in regional enteritis.

Urgent surgical treatment may be called for when there is massive bleeding or an inflammatory condition which cannot be differentiated from acute appendicitis.

Acute cecal diverticulitis and simple ulcers of the cecum constituting the inflammatory "pseudo tumors" of the cecum are rare lesions,[1] not often diagnosed correctly before operation and sometimes with difficulty at the time of operation. Preoperatively, they are usually mistaken for acute appendicitis, and when the abdomen is explored as an emergency, the surgeon is confronted with the finding of a pseudo tumor produced by the surrounding inflammatory process (Fig 1).

Despite the development of a wide variety of sensitive laboratory tests and technical advances in routine roentgenographic examination, localizing the site of gastrointestinal hemorrhage often remains an enigma even at laparotomy.[2] Very frequently the bleeding site is not identified by routine barium x-ray studies.[3] Other means of diagnosing gastrointestinal hemorrhage have been used, such as the fluorescent string test and radioactive-tagged red blood cells.[4] Recently, it has been shown that many of these previously elusive bleeding sites can be demon-

*Read at the national meeting of the Southern Society of Clinical Surgeons, Henry Ford Hospital, April 21, 1970.

**General Surgery Division I

Figure 1

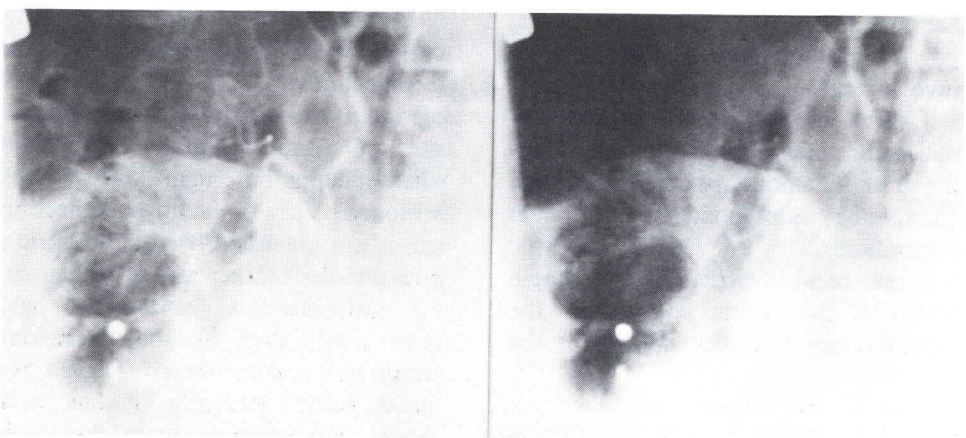

Figure 2

strated by selective visceral arteriography.[2, 5, 6]

Patient Experience

Case No. 1. A 77-year-old man was readmitted following three days of gastrointestinal bleeding. Hemorrhage had occurred on three previous occasions during the past three years. Source of bleeding could not be determined by regular radiographic techniques. By selective mesenteric arteriogram, three tiny venous plexi were revealed in an area of the cecum (x-ray, Figure 2.) They measured about 5 cms in diameter. Also, venous filling from this area appeared greater than normally expected.

When right colectomy was done, gross examination of the cecum revealed a few scarred areas and moderately enlarged ileocolic vessels. When the specimen was opened, the surgeon described a red spot on the lateral wall of the cecum. However, microscopic diagnosis was unremarkable.

The patient has not bled again in the past four years.

Case No. 2. A 62-year-old man had a past history of at least three documented episodes of gastrointestinal bleeding. There had been a previous operation for gastrointestinal bleeding from suspected bleeding ulcer, and a vagotomy and pyloroplasty. Routine gastrointestinal x-rays were negative during recent episodes of bleeding. An intra-arterial mesenteric study was done during the third hemorrhage. Changes were seen compatible

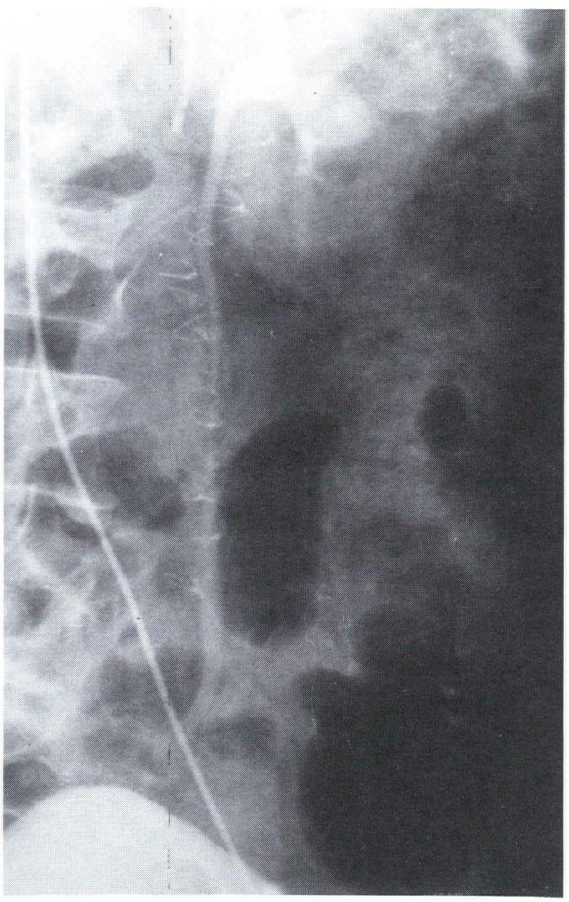

Figure 3

with arterial venous malformation of the cecum (x-ray, Figure 3).

The patient underwent right colectomy. Although gross examination was unremarkable, microscopic examination showed the wall of the veins to be thickened with sclerotic changes. These changes suggested to the pathologist an arterial venous malformation of the cecum (microphotograph; Figure 4).

This patient has been followed for two years with no recurrence of bleeding.

Case No. 3. A 72-year-old male had been studied for chronic iron deficency anemia lasting at least one year. His past history included an exploratory laparotomy in 1963 for an episode of gastrointestinal bleeding. At that time the patient was told his vagus nerves were cut. All gastrointestinal x-ray studies were within normal limits.

Several vessels were seen in the mid-ascending colon in the superior mesenteric arteriogram and there was rapid venous shunting through this area, which measured approximately 5 x 5 cms and was located above the ileocecal valve. An interpretation of carcinoma of the mid right colon (x-ray, Figure 5) led to a right colectomy. The specimen was grossly unremarkable. Under microscopic examination, many blood vessels were seen in the submucosa. There was close association between veins and arteries, but no communication was demonstrated. Luminal vessels were abnormally large and the walls were thinned out. All these features suggested a vascular malformation.

The patient had an uneventful recovery and has been followed for a year without any further bleeding episodes.

Case No. 4. A 60-year-old male had had gastrointestinal bleeding for 18 years. Many radiologic examinations had been negative. He was also known to have severe congestive heart failure. A selective visceral arteriogram was suggestive of hemangioma of the cecum of the arterio-venous variety.

On gross inspection at the time of surgery, there was a thick area in the postero-lateral wall of the cecum and the vessels appeared engorged. No further exploration was done and a modified right colectomy was accomplished. The gross pathological examination showed only a 3 mm area that was slightly depressed, and somewhat reddish-blue in color. Microscopic examination failed to reveal anything comparable to an arterio-venous malformation.

The patient lived for two years without any further gastrointestinal bleeding and finally succumbed of heart disease.

Case No. 5. A 75-year-old woman had

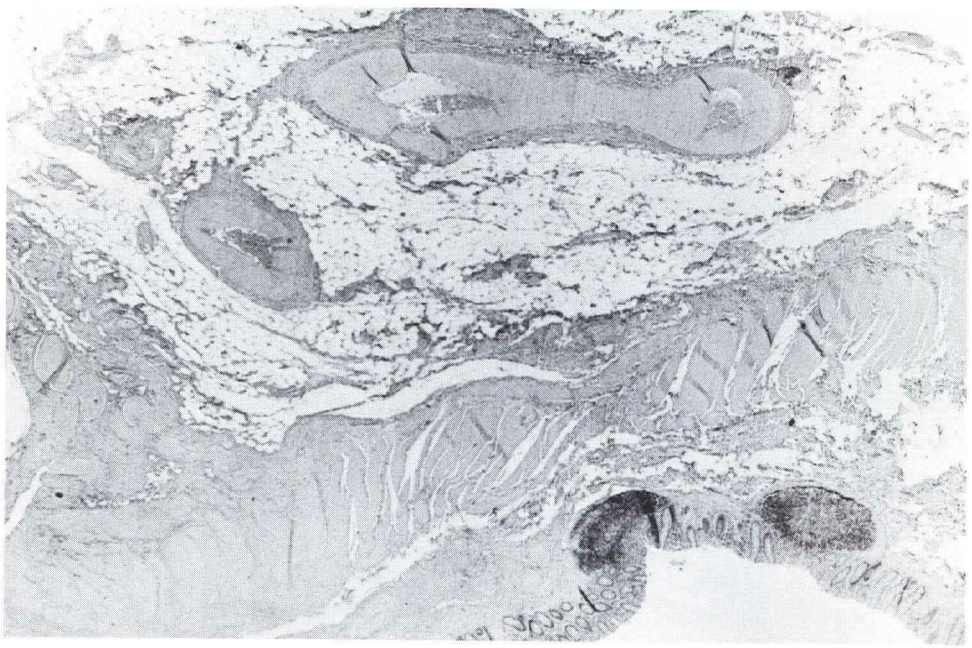

Figure 4

been quite well up to five months prior to being admitted to the hospital for pain located primarily in the lower abdomen. Two months later, she was admitted to another hospital with massive gastrointestinal bleeding which required transfusion of ten units of blood. Barium contrast studies done by that hospital were normal.

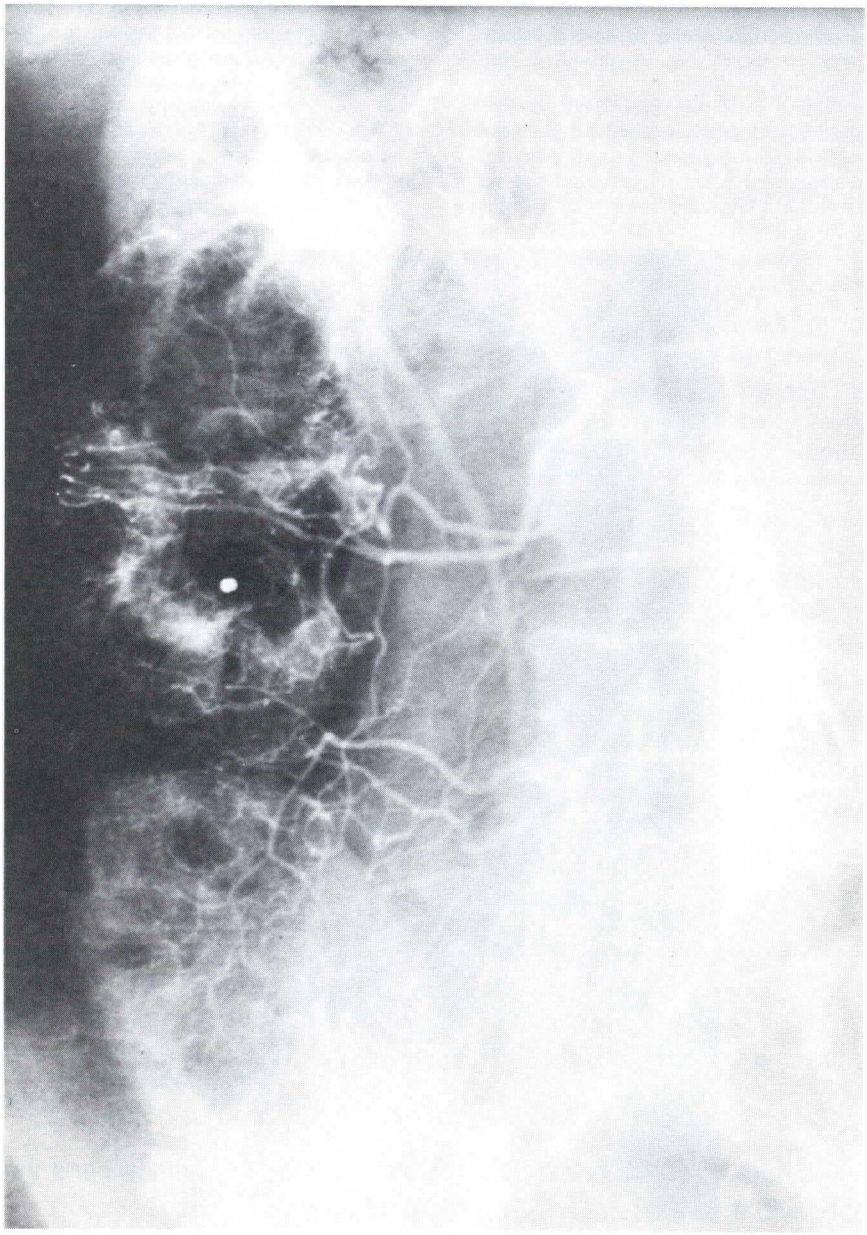

Figure 5

She again was admitted to Henry Ford Hospital for anemia and melena. Her physical examination was essentially negative. A questionable lesion in the hepative flexure was seen in the barium enema. Repeat barium enema was negative. A selective arteriogram indicated the possibility of an A-V malformation in the cecal area. Gross examination of the right colon following laparotomy was unremarkable. A modified right colectomy was performed.

After doing well for three days, the patient suddenly developed a spiked temperature with a low urinary output. She improved somewhat after rehydration and her fever lowered. Her abdomen was painless and not distended. Two days later, she had acute heart failure and cardiac arrest. All resuscitative measures were unsuccessful. An autopsy was not obtained.

Pathologic examination of the specimen showed diverticulosis, with no site of bleeding identified.

Case No. 6. Approximately 1½ years previous to the present admission, this 76-year-old woman had an episode of upper gastrointestinal bleeding at another hospital. Antrectomy and vagotomy were necessary. After doing well for a few months, her hemoglobin count became low and chronic melena was found. Upper and lower GI x-ray studies were essentially negative. The

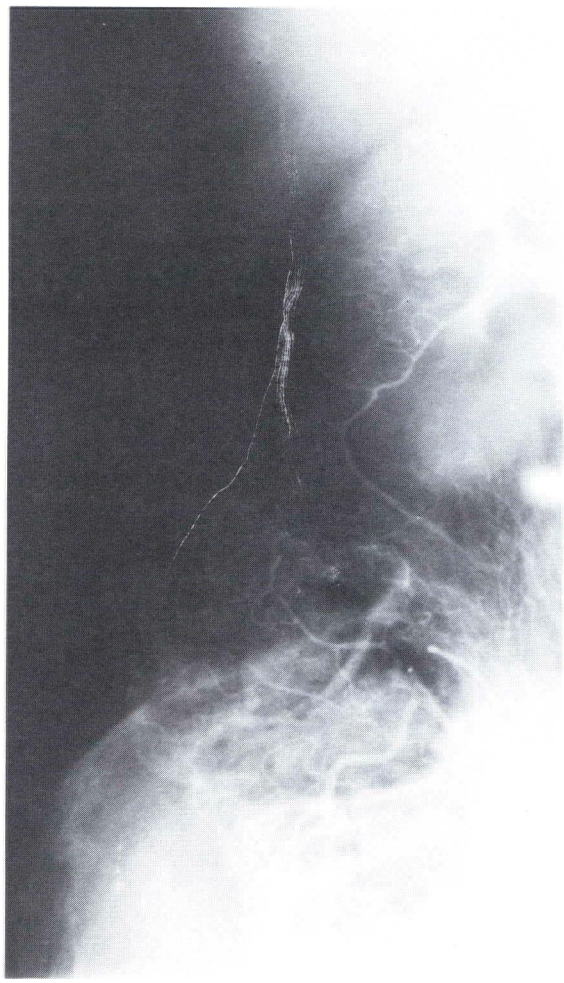

Figure 6

patient was discharged and placed on oral iron medication.

Six months later, she was again found to be anemic, with melena. Gastroscopy and repeat GI x-ray studies failed to reveal the cause for this bleeding. A selective visceral arteriogram disclosed the possibility of an A-V malformation of the cecum (Figure 6). The patient underwent a right colectomy, and gross findings were essentially negative. The report from the pathologist failed to identify the site of bleeding.

Two weeks after the colectomy, the patient had a massive episode of upper GI bleeding and required re-resection of more of her stomach for hemorrhagic gastritis. She recovered.

Discussion

During the past two decades, there has been general recognition that diverticular disease of the colon may be complicated by massive hemorrhage.[7-9] In most such cases, the diagnosis of bleeding from diverticula is made by exclusion, usually from the sigmoid or descending colon. In most instances, the bleeding ceases spontaneously since it originates from ulceration of a small vessel in the diverticulum[8] or, more rarely, from a polyp located therein. Diverticulitis of the cecum is infrequent compared to that occurring in the sigmoid and descending colon (Table I).

Diverticula of the colon are usually in close approximation to blood vessels which are rather large in many instances, especially the veins.[8] Bleeding usually occurs in association with some degree of inflammation but it may occur in apparently uncomplicated cases of diverticulosis. Massive bleeding to the extent that transfusion is required occurs in about 4% of patients with diverticulosis.[7] Only a few need emergency surgery. It is imperative, nevertheless, to determine the site of bleeding; if not, total colectomy and ileorectostomy may be the only way of stopping it.[10] Massive bleeding is rare from a cecal diverticulum whether or not associated with granuloma formation. Raffensperger,[1] reporting on 25 cases of cecal diverticula, found melena in only three of the cases. In no case was there mention of bright red bleeding from the rectum being an indication for

TABLE I

INCIDENCE OF CECAL DIVERTICULAE

Anderson (Mayo Clinic 1947)	9 among 700 cases of surgically-treated diverticulitis
Greaney and Snyder (1957)	14 in a review of 6,781 emergency laparotomies
Rodkey and Hermann (1961)	269 left colon diverticulae and 14 right colon diverticulae in a 20-year review of diverticular disease

emergency surgery. Maier et al[8] reported in 1968 on two cases of cecal diverticulitis complicated by hemorrhage. Emergency surgery was required for these patients.

Bleeding is by far the most common presenting symptom of hemangiomatosis of the intestine.[11,12] Vascular malformations of the intestine are rare (Table II), but are sometimes a source of brisk gastrointestinal bleeding. This ranges in intensity from exsanguinating hemorrhage to microscopic bleeding resulting in secondary anemia. Although there would seem to be a correlation between the occurrence of hemorrhage and an intact mucosa, there have been cases in which an apparently unabraided mucous membrane was found. The diagnosis has rarely been made preoperatively.

Patients with gastrointestinal hemorrhage often present the clinician with a difficult and urgent diagnostic problem. When bleeding is massive, immediate diagnosis and pinpoint localization of the site of bleeding may mean the difference between life and death. A clinical history and physical examination may be revealing or misleading. It has been found that the presence of phleboliths in a plain film of the abdomen is quite common with hemangiomas of the small bowel, especially of the cavernous type.[11] The correct preoperative diagnosis has been made at least four times in this way.[11] The presence of angiomatous lesions of the skin suggests the probability that the intestinal bleeding is caused by similar lesions.[12] Hemangiomata of the small intestine is not uncommon. Although Rissier[11] found arteriovenous malformations of the cecal area in three cases out of 116 reports he reviewed in the literature, such cases are now being reported more frequently because of improved diagnostic techniques.

The prognosis of patients with intestinal hemangiomata is guarded and

TABLE II

INCIDENCE OF VASCULAR MALFORMATION OF THE INTESTINE

Gentry (Mayo Clinic 1949)	106 hemangiomata of the intestines (in a review of 1,400,000 case records)
Ackermann (1937)	3 intestinal hemangiomata in 1200 autopsy records
Rissier (Basle Univ. 1959)	6 cases of cecal hemangiomata among 116 cases of vascular malformation of the intestines

since the therapy of choice is surgical resection, it is imperative to determine the precise bleeding site.

It has been demonstrated in animal experiments that a bleeding rate of from ½ cc to 6 ml per minute can be detected roentgenographically by means of extravasation of contrast material into the bowel lumen. Reports[2,5-6] on the use of selective visceral arteriography to localize bleeding sites in various organs have been appearing in the past few years. Sometimes extravasation cannot be shown, yet pooling or rapid venous filling can indirectly show the source of the hemorrhage. Often this is later confirmed histologically as being limited to the area demonstrated on arteriography. At operation, gross findings may be minimal. Also, as described in our cases and reported by others, microscopic findings may or may not be compatible with arterio-venous malformations. Once the decision has been made, it is very important to proceed with the resection of the portion of bowel diagnosed by the arteriogram to have the lesion.

Conclusions

Vascular malformation of the cecum should be considered in the differential diagnosis of patients with unexplained intestinal bleeding, When a careful history, a meticulous physical examination, and the usual diagnostic methods fail, selective angiography may be quite helpful in locating the site of hemorrhage. Since it appears that surgical resection is the therapy of choice, the precise localization of the bleeding point is important to facilitate its preoperative localization and allow more accurate surgical management.

Unfortunately, the experience so far accumulated and reported in the literature is not extensive enough to establish the frequency of single bleeding episodes caused by vascular lesions in the cecum and small intestine. The fact that we resort to selective angiography only in cases of massive or recurrent bleeding may be partially responsible for this lack of information. It will probably become known when and if selective mesenteric angiography becomes part of the clinical investigation of all cases of gastrointestinal bleeding.

As the number of reported cases increases, we will also be able to learn how often arteriography fails to identify these lesions.

Acknowledgment

The authors express their appreciation to Dr. R. J. Priest (Division of Gastroenterology) and Dr. R. L. Rian (Department of Radiology) for their cooperation in the diagnosis of these patients.

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