Colonic Hemorrhage From a Solitary Minute Ulcer
Report of Three Cases

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Three patients with massive lower gastrointestinal bleeding are reported. In all cases, the bleeding source was localized by emergency selective mesenteric angiography. The histologic lesion found in the resected specimen consisted of a minute mucosal ulcer with an abnormally large eroded submucosal artery without evidence of true angiodysplastic changes. The clinicopathological picture is similar to the rare solitary stomach ulceration, described as "Exulceratio simplex Dieulafoy."

Vascular malformation of the bowel has been reported as a cause of severe gastrointestinal bleeding with increasing frequency (1–4). Angiodysplasia is a vascular malformation, almost always localized in the cecum and ascending colon (5). The abnormality consists of minute clusters of dilated vessels, mostly veins, in the mucosa and submucosa of the right colon. These vascular malformations have been referred to under a variety of terms such as "arteriovenous malformation" (6), "teleangectasia" (7), or "angioma" (8). Three patients with massive lower gastrointestinal hemorrhage, resulting from a solitary arterial abnormality in the submucosal layer of the ascending colon which was different from angiodysplastic malformations, are reported.

Case Reports

Case 1

A 59-yr-old man with no history of previous gastrointestinal bleeding was admitted with massive rectal hemorrhage and profound shock with an estimated blood loss of 7 L. Emergency upper gastrointestinal endoscopy was negative. Systemic vasopressin infusion* failed to control the hemorrhage. Emergency fiber rectosigmoidoscopy was not conclusive, because rapidly accumulating blood from above prevented good visualization. Angiography showed contrast medium accumulating in the ascending colonic lumen from a branch of the right colic artery without evidence of an intrinsic vascular abnormality. At immediate laparotomy, no bleeding source could be identified. A right hemicolec-tomy was performed on the basis of the angiographic findings. A minute epithelial defect (2 mm in diameter) in the ascending colon was the only abnormal finding in the resected specimen (Figure 1A).

Histologically, an acute ulceration extending to the uppermost part of the submucosa was detected. An abnor-mally thick-walled ruptured artery was located within the ulceration just below the muscularis mucosae (Figures 1B and 1C). This large-caliber vessel showed no appreciable changes of atherosclerosis. In serial sections no other abnormality was found.

Case 2

A 74-yr-old male patient was admitted in circulatory shock after massive rectal bleeding, requiring 5 L of blood in the first 4 h. Upper gastrointestinal endoscopy was negative and subsequent colonoscopy was inconclusive because of persistent bleeding. Systemic vasopressin infusion (identical to case 1) failed to control the bleeding. Angiography demonstrated contrast medium extravasation in the lumen of the ascending colon. An emergency laparotomy was performed.

A small area of induration was detected on careful palpation of the ascending colon. A partial resection of the ascending colon was performed. Histologically, a thick-walled tortuous artery was seen just beneath and parallel to the muscularis mucosae in close proximity to the

* The vasopressin therapy was identical in all 3 cases (initial 20 U bolus, followed by 0.4 U/min).
Figure 1. Gross appearance and photomicrograph of colonic lesion from case 1. A. Part of the gross specimen with a minute solitary reddish lesion (encircled) in an otherwise normal mucosa. B. Low magnification shows the fresh ulceration penetrating the submucosa and eroding the thick-walled artery (arrow) located just below the muscularis mucosae (arrowheads). (Van Gieson-elastic, x 25.) C. Ruptured artery at higher magnification. Apart from the abnormal caliber and the thick wall, the vessel exhibits no pathological changes. (Van Gieson-elastic, x 90.)

Case 3

A 63-yr-old man with a 3-yr history of panmyelopathy of unclear etiology, which manifested clinically solely as a recurrent pyodermia and had not caused an increased bleeding tendency, was admitted in circulatory shock due to acute massive peranal bleeding. He required transfusion of 12 U of whole blood in the first 24 h. Upper gastrointestinal endoscopy was normal and emergency colonoscopy was inconclusive due to the blood-filled colonic lumen. Systemic vasopressin infusion (identical to case 1) led to reduction of the rectal bleeding. Nevertheless, as cessation of bleeding was not achieved, a colonic washout with 3000 ml of Golytely (Braintree Laboratories, Inc., Braintree, Mass.) solution (9) was performed before a further colonoscopy. A minute ulceration at the junction part of the arterial wall were of normal thickness with neither fibrosis nor atherosclerosis.

Figure 2. Photomicrograph of colonic lesion from case 2. A. Histology at low magnification showing a minute ulcer with ruptured artery (arrow). Note reduction of wall thickness and the tiny aneurysm containing embolized material (cholesterol crystals). No pathological changes of the mucosal and submucosal layer. Arrowheads point to the muscularis mucosae. B, remnants of clotted blood. (H & E, x 18.) B. Adjacent to the rupture, the thick-walled tortuous artery runs just beneath the muscularis mucosae (arrowheads) (H & E, x 30.)
of the cecum and ascending colon with an adherent clot was detected. No active bleeding was observed at this site during the procedure. Angiography was performed after further massive peranal bleeding. Contrast medium extravasation in the lumen of the ascending colon was demonstrated. At laparotomy, no lesion was found. A right hemicolectomy was performed. Histologically, the sole finding was a fresh hemorrhagic ulceration with a defect in the wall of an underlying submucosal artery without any features of angiodysplasia.

No further episode of gastrointestinal hemorrhage was documented in all 3 cases.

Discussion

In the cases presented, the source of the massive lower gastrointestinal bleeding was a minute mucosal ulceration in the right colon with erosion of a large submucosal muscular artery. There were no features suggesting a vascular abnormality such as angiodysplasia. The bleeding was massive in each patient. There was no evidence of previous bleeding. The source of bleeding from the gastrointestinal tract was localized by selective superior mesenteric arteriography.

The clinicopathologic picture of the described cases is very similar to the so-called “Exulceratio simplex Dieulafoy” of the stomach, a condition first described in 1898 by the French clinician Georges Dieulafoy (1839–1911) (10). Exulceratio simplex has been found almost exclusively in the stomach (10–15). Two cases of exulceratio-like lesions involving the jejunum have been reported (16).

Exulceratio simplex is characterized morphologically by a small solitary mucosal defect with erosion of an underlying submucosal large-caliber artery (11–15). Histologically, this ulceration extends not deeper than the upper submucosal layer (10). The large-caliber submucosal artery (diameter ≤2 mm) in the region of the ulceration is tortuous and curved toward the mucosal membrane, and is eroded at the apex of its curve (11). These vessels have the typical structure of larger muscular arteries and are similar to the supplying subserosal arteries; thus the term “caliber persistence” has been used (17). These vessels, morphologically normal otherwise, may also show atheromatous changes (13) (case 2). The described anomaly is totally different from the vascular malformation encountered in angiodysplasia.

The nature of exulceratio simplex-like lesions is unclear. It seems to be a localized vascular anomaly, as similar vascular changes are not found at other sites of the gastrointestinal tract (13,16). As exulceratio simplex-like lesions can be found in young patients without atherosclerosis (16), the inference is that this condition is a primary vascular anomaly and not an acquired abnormality due to atherosclerosis.

A likely pathogenesis is that the strong pulsations of a large tortuous submucosal artery may mechanically damage the overlying mucosa and induce ulceration. The exposed arterial wall may consequently be eroded by bowel contents. This is supported by the fact that there are no other ulcerations than the one directly over this enlarged artery. The ulceration seems to be secondary to the isolated vascular anomaly. This is supported by the fact that no other mucosal or vascular abnormalities were found in our resected specimens. The resultant bleeding seems always to be massive, which may be due to the fact that the defect is tangential to the lumen and effective hemostasis by vascular contraction or thrombosis is unlikely (11).

Conservative management by systemic vasopressin infusion has not been successful in our 3 cases, which is somewhat surprising as diverticular bleeding from arteries of a similar size responds well to this treatment (18,19).

In summary, 3 patients with massive lower gastrointestinal hemorrhage from a minute ulcer and with erosion of an underlying abnormally large submucosal artery as the only pathological changes are reported. Lesions, such as described in this paper, may in the past have been the cause of severe lower gastrointestinal hemorrhage of undetermined origin.

The frequency of such large-caliber submucosal arteries can only be established by further evaluation of specimens obtained during colon resection.

References

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electrolyte absorption or secretion. Gastroenterology 1980; 78:991.


