Gastric Arteriovenous Malformation with Characteristic Endoscopic Findings

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Abstract:
Gastric arteriovenous malformation (AVM) is an uncommon cause of upper gastrointestinal bleeding, and the endoscopic findings are unclear. We herein describe a case of gastric AVM in a 28-year-old man. Esophagogastroduodenoscopy showed a Dieulafoy lesion surrounded by a red mucosa with a sharp margin, which implied blood vessel malformation. Computed tomography angiography and conventional angiography revealed aggregated vessels on the greater curvature. Partial gastrectomy was performed, with no recurrent bleeding postoperatively. The histopathological diagnosis was AVM. We conclude that gastric AVM should be considered in the differential diagnosis of patients who present with a Dieulafoy lesion surrounded by a red mucosa.

Key words: gastric arteriovenous malformation, red mucosa, endoscopy, Dieulafoy lesion

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Introduction
Gastric arteriovenous malformation (AVM) is an uncommon cause of upper gastrointestinal bleeding. However, it can be fatal and almost always requires surgical treatment. The endoscopic findings of gastric AVM are unclear and have only rarely been reported (1). We herein report a rare case of gastric AVM with characteristic findings on gastroduodenoscopy.

Case Report
A 28-year-old Asian man who had undergone endoscopic hemostasis three times over the previous six years presented to our hospital with epigastric pain. Physical assessment revealed normal vital signs, normal conjunctivae, and tarry stool. His past medical history was unremarkable other than three episodes of upper gastrointestinal bleeding. His social and family histories were unremarkable. His laboratory test findings showed mildly elevated blood urea nitrogen; all other values were within the normal range. Emergency esophagogastroduodenoscopy showed a Dieulafoy lesion surrounded by a sharply demarcated red mucosa in the greater curvature of the middle gastric body (Fig. 1A and B). During endoscopy, spurting-type bleeding (Forrest 1a) without any stimulation was seen from the Dieulafoy lesion (Fig. 1C); this lesion was considered to be the likely cause of his bleeding and tarry stool. He was first treated with local epinephrine injection. Despite multiple injections, the bleeding continued. Hemostatic clips were then applied and hemostasis was thereafter achieved.

Computed tomography - angiography and conventional angiography revealed aggregated vessels on the greater curvature (Fig. 2), which were suggestive of gastric AVM. After receiving the patient's informed consent, he requested a second opinion. He underwent partial gastrectomy at an outside hospital because endoscopic treatment could
not be performed due to the thickness of the AVM, as demonstrated by CT, especially after the repeated failure to achieve endoscopic hemostasis. The patient desired partial gastrectomy despite the risk of recurrence in the remaining stomach. The histopathological diagnosis was AVM (Fig. 3). There has been no recurrent hemorrhage during the 6 months of postoperative follow-up.

**Discussion**

We herein describe a rare case of gastric AVM with characteristic endoscopic findings, consisting of a Dieulafoy lesion surrounded by a sharp margin of red mucosa on the greater curvature. This case demonstrates two significant features: first, gastric AVM can present as a Dieulafoy lesion; second, the presence of a Dieulafoy lesion, especially when surrounded by a red mucosa, might suggest the presence of gastric AVM, except when located on the lesser curvature.

Gastric AVMs have only very rarely been reported; they represent 1.4% of all intestinal AVMs (2). Case reports of gastric AVM were published beginning in the 1880s, when it was reported to cause massive upper gastrointestinal bleeding and death (3). Since the 1970s, gastric AVM has been diagnosed with endoscopy and it is normally treated surgically (4-7). In the 2000s, successful endoscopic therapy (8, 9) and balloon-occluded retrograde transvenous obliteration (10) were reported. However, reports describing the endoscopic findings of gastric AVM remain relatively rare (Table). Moreto (1) classified 47 cases of gastric and duodenum AVM into three groups according to endoscopic findings: bright red lesions with a frond-like margin, a telangiectatic form, and a submucosal nodular form. However, Dieulafoy lesions were not mentioned. With the exception of Moreto’s publication, Table summarizes the published case reports of gastric AVM reporting the endoscopic findings in English retrieved from a PubMed search, none of which included any Dieulafoy lesions. Outside of the English literature, two cases of Dieulafoy lesion associated with gastric AVM have been reported in the Japanese literature (11, 12). Our patient might thus be the first reported case of gastric AVM with a Dieulafoy lesion in the English literature.

A Dieulafoy lesion is an important cause of acute gastrointestinal bleeding because of its propensity to cause massive hemorrhaging. The proximal stomach, in particular within 6 cm of the gastroesophageal junction and along the lesser curvature, is the most common gastric location, accounting for approximately 75% of all gastric lesions (13).
The blood supply to this region comes directly from the arterial chain because the arterial plexus is absent in this area (14). While the cause of Dieulafoy lesions is unclear, the vascular steal phenomenon is hypothesized to play a role: namely, an enhanced blood flow through an enlarged artery may cause hypoperfusion and ischemia of the overlying mucosa (15), and this pulsation thus transmits mechanical pressure, which may lead to the development of Dieulafoy lesions. We think this hypothesis supports AVM as a cause of Dieulafoy lesions by the same mechanism. Thus, the presence of a Dieulafoy lesion, except when on the lesser curvature, might suggested the presence of AVM.

A sharp margin of the red mucosa surrounding the ulcer, as in this case, could further suggest AVM, as Moreto (1) reported similar endoscopic findings with gastric AVM: namely, bright red lesions with a frond-like margin.

Scant data on gastric AVM are currently available, and classifying the endoscopic findings associated with gastric

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**Figure 3.** Pathological image of gastric arteriovenous malformation. A: Surgical specimen, B: Cut specimen; C, D are magnified images of the red circle and E, F are magnified image of the yellow circle, C: Dilated, tortuous submucosal vessels (black arrows) and large vessel showing arteriovenous malformation (AVM) (blue arrow). Hematoxylin and Eosin (H&E) staining; magnification×12.5, D: Magnified image of C (blue arrow) shows large vessels with the thickness of the smooth muscle differing on the left and right. This implies arterial (thick smooth muscle) vessels joining venous (narrow smooth muscle) vessels, which indicates AVM. Elastica Van Gieson’s stain; magnification×40, E: Ruptured vessel (blue arrow) with overlying small mucosal ulceration (black arrows). H&E staining; magnification×40, F: Magnified image of E (blue arrow) shows a ruptured vessel with irregular elastic fiber. Elastica Van Gieson’s stain; magnification×200.
The authors state that they have no Conflict of Interest (COI).

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In conclusion, we herein reported a rare case of gastric AVM with characteristic endoscopic findings, consisting of a Dieulafoy lesion surrounded by a sharp margin of a red mucosa.

| Case No. | Year | Age | Sex | Location of AVM | Endoscopic Findings | Diagnostic methods | Therapy | Result | Reference |
|---------|------|-----|-----|-----------------|---------------------|-------------------|---------|--------|-----------|
| 1       | 1974 | 44  | M   | Greater curve   | No definite vascular lesion | Angiography | OPE | Alive | (4) |
| 2       | 1975 | 48  | F   | Posterior wall  | Pin point defect | Pathological | OPE | Alive | (5) |
| 3       | 1976 | 74  | F   | Greater curvature in the cardia | Massive blood, nothing about AVM | Pathological | OPE | Alive | (6) |
| 4       | 1977 | 16  | F   | Cardia          | Mucosal tear       | Angiography | OPE | Alive | (7) |
| 5       | 1980 | 64  | F   | Fundus          | Small area of redness | Autopsy |     | Dead  | (16) |
| 6       | 1981 | 67  | F   | Fundus          | Polyp or leiomyoma   | Pathological | OPE | Alive | (17) |
| 7       | 1981 | 21  | M   | Cardia          | Vascular malformation | Pathological | OPE | Alive | (17) |
| 8       | 1981 | 64  | F   | Fundus          | Enlarged mucosal folds | Angiography | Angiography | Alive | (18) |
| 9       | 1983 | 56  | F   | Posterior wall  | Nothing abnormal | Pathological | OPE | Alive | (19) |
| 10      | 1985 | 66  | M   | Posterior wall  | Normal              | Pathological | OPE | Alive | (20) |
| 11      | 1986 | 60  | M   | Polyp          | Polyp               | Pathological | OPE | Alive | (21) |
| 12      | 1994 | 69  | F   | Posterior wall  | SMT, telangiectasia | Pathological | OPE | Alive | (22) |
| 13      | 2002 | 38  | F   | Greater curve   | Small bulge         | Doppler EUS | Endoscopic therapy | Alive | (8) |
| 14      | 2007 | 50  | M   | Greater curve   | SMT                 | Pathological | OPE | Alive | (23) |
| 15      | 2009 | 30  | F   | Greater curve   | Gastric ulcer       | Pathological | OPE | Alive | (24) |
| 16      | 2011 | 14  | M   | Fundus          | Normal              | Pathological | OPE | Alive | (25) |
| 17      | 2016 | 85  | M   | Greater curve   | SMT                 | Pathological | OPE | Alive | (26) |
| 18      | 2016 | 87  | M   | antrum          | SMT                 | Pathological | ESD | Alive | (9) |

SMT: submucosal tumor, OPE: operation, EUS: endoscopic ultrasonography, ESD: endoscopic submucosal dissection
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