

[CASE REPORT]

Gastric Arteriovenous Malformation with Characteristic Endoscopic Findings

Yoji Hirayama¹, Chihiro Takai², Kai Korekawa¹, Masahiro Saito¹, Asuka Akasaka¹,
Hitoshi Nagasawa¹, Wataru Iwai³, Hisashi Hachimori¹, Masahiro Satoh¹,
Nobuhisa Yajima⁴ and Motoji Oki¹

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

(Intern Med 57: 2341-2345, 2018)

(DOI: 10.2169/internalmedicine.0003-17)

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 gastro-duodenoscopy.

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

¹Department of Gastroenterology, Endoscopy and Chemotherapy, Hachinohe City Hospital, Japan, ²Department of General Internal Medicine, Mito Kyodo General Hospital, Japan, ³Department of Gastroenterology, Miyagi Cancer Center, Japan and ⁴Department of Pathology, Hachinohe City Hospital, Japan

Received: August 1, 2017; Accepted: December 21, 2017; Advance Publication by J-STAGE: March 9, 2018

Correspondence to Dr. Yoji Hirayama, hirayamayoji@gmail.com

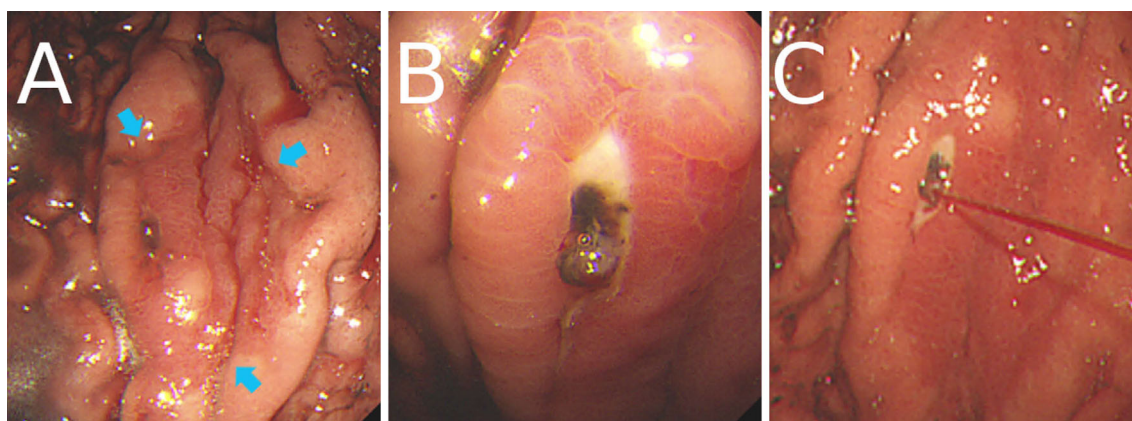


Figure 1. Esophagogastroduodenoscopy on day 1. A: Dieulafoy ulcer surrounded by a sharp margin (blue arrows) of red mucosa on the greater curvature of the middle gastric body, B: Extended image of the Dieulafoy ulcer, C: Spurting bleeding (Forrest 1a) was present without any stimulation.

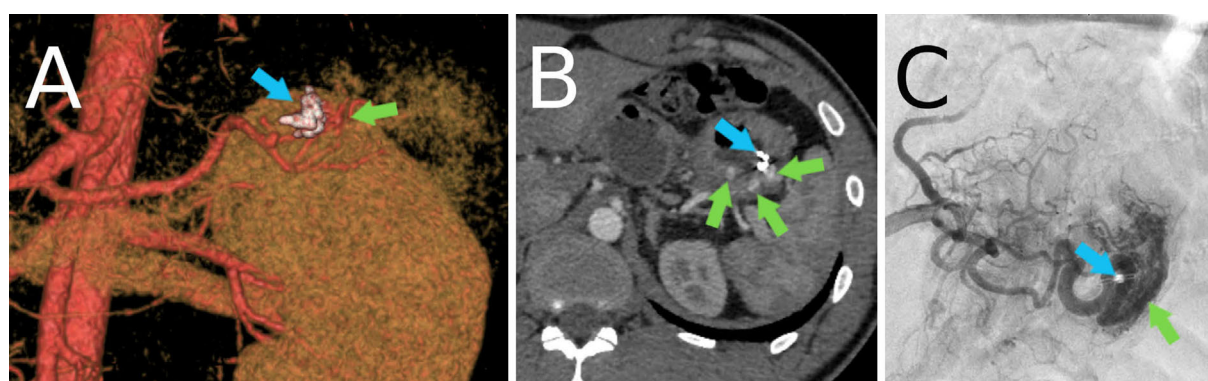


Figure 2. Imaging studies show enhancing vascular anomaly (green arrow) involving the greater curvature near the clips (blue arrow). A: 3D-CT Angiography, B: CT, C: Angiography.

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).

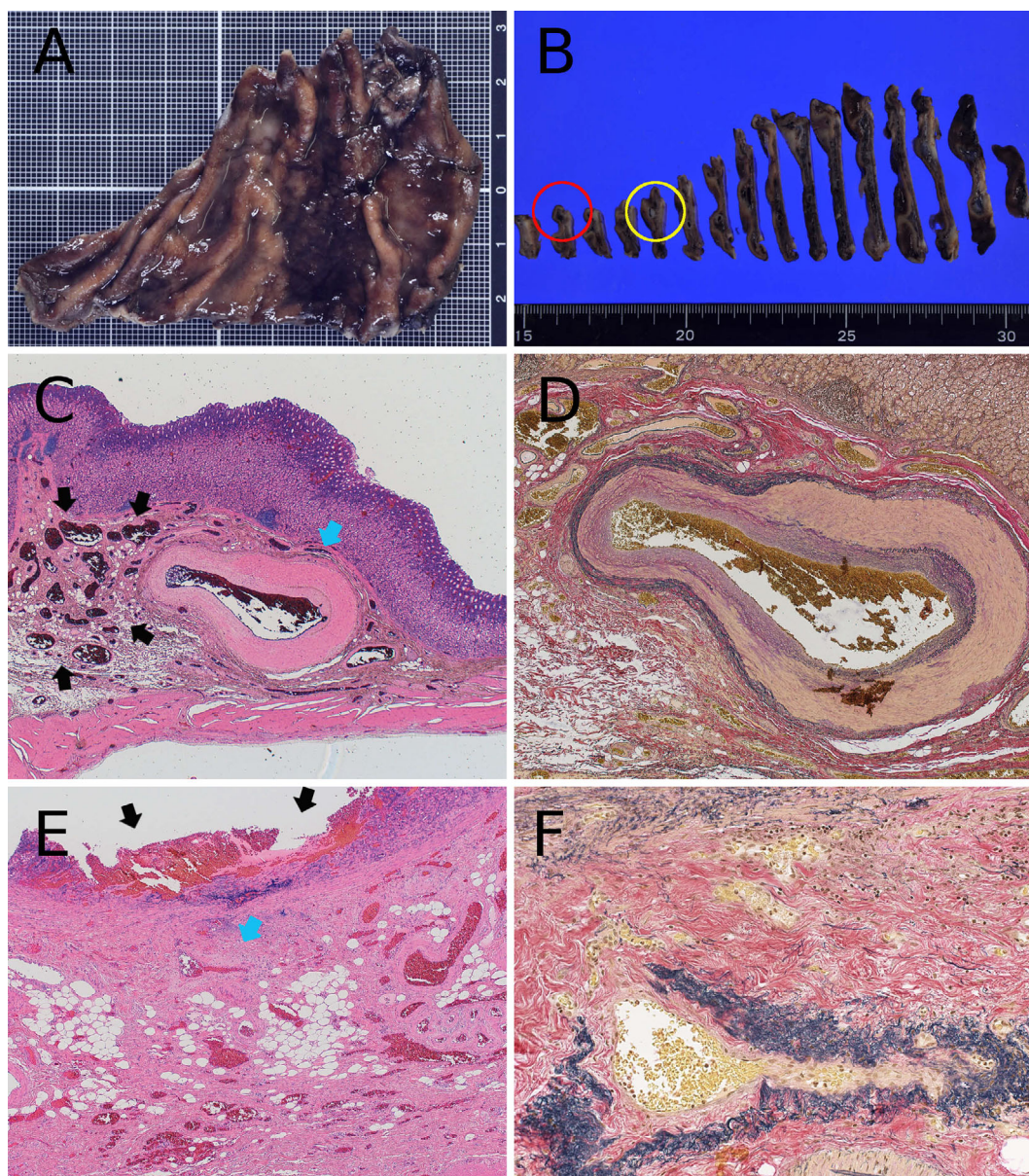


Figure 3. Pathological image of gastric arteriovenous malformation. A: Surgical specimen, B: Cut specimen; C, D are magnified images of the red circle and E, F are magnified image of the yellow circle, C: Dilated, tortuous submucosal vessels (black arrows) and large vessel showing arteriovenous malformation (AVM) (blue arrow). Hematoxylin and Eosin (H&E) staining; magnification $\times 12.5$, 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 $\times 40$, E: Ruptured vessel (blue arrow) with overlying small mucosal ulceration (black arrows). H&E staining; magnification $\times 40$, F: Magnified image of E (blue arrow) shows a ruptured vessel with irregular elastic fiber. Elastica Van Gieson's stain; magnification $\times 200$.

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

Table. Cases of Gastric Arteriovenous Malformation Diagnosed with Endoscopy.

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	Dead	(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	Gastro-esophageal junction on the lesser curvature	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

AVM may be useful to determine the pathogenesis and thereby choose the most efficacious therapeutic approach. Further reports on gastric AVM are thus needed in order to accumulate detailed data on the associated endoscopic findings.

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.

The authors state that they have no Conflict of Interest (COI).

References

- Moreto M, Figa M, Ojembarrena E, Zaballa M. Vascular malformations of the stomach and duodenum: an endoscopic classification. *Endoscopy* **18**: 227-229, 1986.
- Meyer CT, Troncale FJ, Galloway S, Sheahan DG. Arteriovenous malformations of the bowel: an analysis of 22 cases and a review of the literature. *Medicine* **60**: 36-48, 1981.
- Gallard T. Anéurysmas miliaries de l'estomac, donnant lieu à des hématémèses mortelles. *Bull Soc Med Hôp Paris* **1**: 84, 1884 (in French).
- Pastershank SP, Chappell EW, Buchan DJ, Chow KC. Arteriovenous malformation of the stomach. *Can Med Assoc J* **110**: 57, 1974.
- Richter RM. Massive gastric hemorrhage from submucosal arterial malformation. *Am J Gastroenterol* **64**: 324-326, 1975.
- Chandler KE, Cheek RC. Ruptured submucosal gastric artery microaneurysm. *Am Surg* **42**: 421-426, 1976.
- Gough MH. Submucosal arterial malformation of the stomach as the probable cause of recurrent severe haematemesis in a 16-year-old girl. *Br J Surg* **64**: 522-524, 1977.
- Pedersen FM, Vilmann P, Bytzer P. Gastric arteriovenous malformation: Doppler EUS-guided diagnosis and therapy. *Gastrointest Endoscop* **55**: 597-599, 2002.
- Kojima Y, Takeuchi T, Egashira Y, et al. Endoscopic submucosal dissection for gastric arteriovenous malformation. *Int Med* **55**: 3221-3223, 2016.
- Hanshing CE, Marquardt JP, Sutton DM, et al. Balloon-occluded retrograde transvenous obliteration of a gastric vascular malformation: an innovative approach to treatment of a rare condition. *Cardiovasc Interv Radiol* **40**: 310-314, 2017.
- Kimura M, Tanaka T, Tsuji M, et al. A case of Dieulafoy's ulcer due to A-V malformation demonstrated by endoscopic ultrasonography. *Gastroenterol Endosc* **31**: 2454-2462, 1989 (in Japanese, Abstract in English).
- Idogawa A, Nagashima A, Arakawa T, Yamataga S, Fujisaki M, Sendo K. A case of gastric arteriovenous malformation observed endoscopically before and after bleeding. *Gastroenterol Endosc* **34**: 117-122, 1992 (in Japanese, Abstract in English).
- Van Zanten SV, Bartelsman JF, Schipper ME, et al. Recurrent massive haematemesis from Dieulafoy vascular malformations - a review of 101 cases. *Gut* **27**: 213-222, 1986.
- Barlow TE, Bentley FH, Walder DN. Arteries, veins, and arteriovenous anastomoses in the human stomach. *Surg Gyn Obstet* **93**: 657, 1951.
- Juler GL, Labitzke HG, Lamb R, Allen R. The pathogenesis of

- Dieulafoy's gastric erosion. *Am J Gastroenterol* **79**: 195-200, 1984.
16. Mohammad A, O'Neal RM. Fatal hemorrhage from a ruptured aneurysm of a submucosal gastric artery. *South Med J* **73**: 952-953, 1980.
 17. Lewis JW, Mason EE, Jochimsen PR. Vascular malformations of the stomach and duodenum. *Surg Gyn Obstet* **153**: 225, 1981.
 18. Sellu DP, Fagan E, Allison DJ, Wood CB. Arteriovenous malformation of the stomach treated by embolization. *Br J Surg* **68**: 39-40, 1981.
 19. Okada M, Iida M, Fuchigami T, et al. Submucosal arterial malformation of the stomach diagnosed endoscopically. *Gastrointest Endoscop* **29**: 30-31, 1983.
 20. Teng PK, Cho S. Massive gastric hemorrhage from the rupture of an arterial malformation. *NY State J Med* **85**: 112, 1985.
 21. Tio TL, Tytgat GNJ. Endoscopic ultrasonography of an arteriovenous malformation in a gastric polyp. *Endoscopy* **18**: 156-158, 1986.
 22. Kono K, Sekikawa T, Iino H, Ogawara T, Matsumoto Y. A case of arteriovenous malformation in the submucosal layer of the stomach. *J Gastroenterol* **29**: 340-343, 1994.
 23. Araki T, Wakabayashi T. Gastric arteriovenous malformation. *Int Med* **46**: 915-916, 2007.
 24. Khan MI, Baqai MT, Baqai MF, Mufti N. Exsanguinating upper GI bleeds due to unusual arteriovenous malformation (AVM) of stomach and spleen: a case report. *World J Emerg Surg* **4**: 15, 2009.
 25. Latar NH, Phang KS, Yaakub JA, Muhammad R. Arteriovenous malformation of the stomach: a rare cause of upper gastrointestinal bleeding. *Med J Malaysia* **66**: 142-143, 2011.
 26. Hotta M, Yamamoto K, Cho K, Takao Y, Fukuoka T, Uchida E. Stomach arteriovenous malformation resected by laparoscopy-assisted surgery: a case report. *Asian J Endoscop Surg* **9**: 135-137, 2016.

The Internal Medicine is an Open Access article distributed under the Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License. To view the details of this license, please visit (<https://creativecommons.org/licenses/by-nc-nd/4.0/>).