

[CASE REPORT]

Massive Bleeding from a Small Intestinal Submucosal Tumor-like Arterial Malformation: An Unclassifiable Vascular Lesion Revealed by a Detailed Pathological Evaluation

Kazuhiro Ota¹, Junki Noda¹, Hiroshi Akutagawa², Noriaki Sugawara¹, Kei Nakazawa¹, Shinya Nishida¹, Yuki Hirata¹, Satoshi Harada¹, Wataru Osumi³, Yuichi Kojima¹, Sadaharu Nouda¹, Toshihisa Takeuchi¹, Yoshinobu Hirose² and Kazuhide Higuchi¹

Abstract:

A 53-year-old woman presented with repeated copious bloody stool. Small bowel capsule endoscopy revealed a submucosal tumor (SMT)-like lesion, with erosion of the surface, in the first third of the small bowel. Balloon-assisted small intestinal endoscopy also revealed a pulsatile SMT-like lesion with an exposed vessel on the surface. This unknown lesion was surgically resected. The histopathological findings of the resected SMT-like lesion showed a dilated artery with thrombosis blockage and recanalization. Since this case could not be classified as any of the small intestinal vascular lesion patterns endoscopically, its classification will require the accumulation of further cases.

Key words: obscure gastrointestinal bleeding, capsule endoscopy, small intestinal bleeding

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Introduction

Histopathologically, small intestinal vascular lesions have been classified as angioectasia, Dieulafoy's lesion, and arteriovenous malformation. The Yano-Yamamoto classification is widely used for the endoscopic classification of small intestinal vascular lesions (Table) (1). It is generally agreed that the Yano-Yamamoto classification types 1a and 1b correspond to angioectasia, types 2a and 2b to Dieulafoy's lesion, and type 3 to arteriovenous malformation; however, type 4 does not correspond to a specific lesion. Some types of small intestinal lesions cause massive arterial bleeding, including arterial vascular lesions (Dieulafoy's lesion and arteriovenous malformation), ulcers, and malignant tumors (gastrointestinal stromal tumor and cancer). Small intestinal arterial bleeding often causes recurrent episodes of sudden massive bloody stool and hemorrhagic shock without ab-

dominal pain and requires a long-term examination to detect its origin. For these reasons, unidentified and unclassified lesions should be further studied.

We herein report a case of massive bleeding from a small intestinal submucosal tumor (SMT)-like arterial malformation.

Case Report

A 53-year-old woman presented with recurrent copious bloody stool and hemorrhagic shock. She underwent dynamic contrast-enhanced computed tomography, emergent esophagogastroduodenoscopy, and total colonoscopy at another hospital; however, the bleeding origin could not be identified. Therefore, she was transferred to our hospital for the further investigation of the small intestinal bleeding. Because small bowel capsule endoscopy (PillCam[®]SB3; Covidien, Dublin, Ireland) revealed a hemispherical SMT-

¹Second Department of Internal Medicine, Osaka Medical College, Japan, ²Department of Pathology, Osaka Medical College, Japan and ³Department of General and Gastroenterological Surgery, Osaka Medical College, Japan

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Correspondence to Dr. Kazuhiro Ota, clash_kaz@yahoo.co.jp

Table. Endoscopic Classification of Small Intestinal Vascular Lesion (Yano-Yamamoto Classification).

Type 1a	Puncture erythema (<1 mm) with or without oozing
Type 1b	Patchy erythema (a few mm) with or without oozing
Type 2a	Puncture lesions (<1 mm) with pulsatile bleeding
Type 2b	Pulsatile red protrusion without surrounding venous dilation
Type 3	Pulsatile red protrusion with surrounding venous dilation
Type 4	Other lesions not classified into any of the above categories

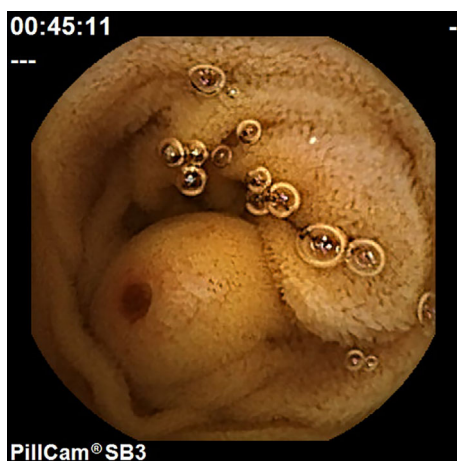


Figure 1. Capsule endoscopic findings (PillCam®SB3; Covidien, Dublin, Ireland) revealed a hemispherical submucosal tumor-like lesion with erosion at the surface in the first third of the small bowel.

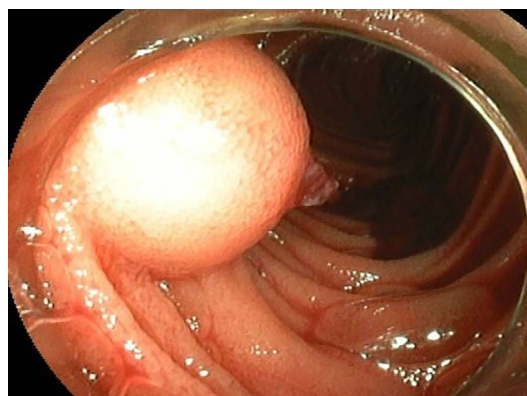


Figure 2. Transoral double-balloon enteroscopy also revealed a pulsatile hemispherical submucosal tumor-like lesion with an exposed vessel at the surface.



Figure 3. Macroscopic findings of the surgically resected specimen. Note the submucosal tumor-like lesion with erosion at the anal side of the markings with clipping and inking.

like lesion with erosion of the surface in the first third of the small bowel, the lesion was identified as the bleeding origin (Fig. 1, Supplementary material). Transoral double-balloon enteroscopy also revealed a pulsatile hemispherical SMT-like lesion with an exposed vessel on the surface, not accompanied by surrounding venous dilatation (Fig. 2).

The lesion was classified as type 4 (unclassifiable) in the Yano-Yamamoto classification (1). However, although gastrointestinal stromal tumor and arteriovenous malformation were included as the differential diagnosis of this lesion, it could not be definitively diagnosed by endoscopic findings and previous studies alone. Furthermore, because the com-

pleteness of endoscopic hemostasis for this unknown lesion remained uncertain, it was surgically resected (Fig. 3). The histopathological findings of the resected SMT-like lesion showed a dilated artery with thrombosis blockage and recanalization. In a high-magnification Elastica van Gieson-stained image, the elastic lamina was confirmed to have no venous component (Fig. 4).

Discussion

In the present case, dynamic contrast-enhanced computed tomography did not reveal the bleeding origin; however, small bowel capsule endoscopy effectively detected the origin and location of bleeding to guide the approach route of balloon-assisted enteroscopy, which did not achieve hemostasis but effectively marked the lesion for surgical resection. The immediate diagnosis of the occult gastrointestinal bleeding based on an efficient strategy is necessary for early hemostasis (2). When deciding on the treatment strategy for bleeding lesions with uncertain endoscopic diagnosis, endoscopic hemostasis should be carefully evaluated (3).

Several previous case reports have been conducted on small intestinal vascular malformations causing obscure gastrointestinal bleeding. Liao et al. reported a vascular malformation in the small intestine that was an SMT-like lesion with erosion at the surface, as detected endoscopically. Their case report's common pathological point is similar to that of our case: thrombosis with thick-walled blood vessels detected histologically (4). Sai et al. considered that an ele-



Figure 4. Microscopic findings showing a dilated artery with thrombosis blockage and recanalization, rather than a tumor. In the high-magnification image on Elastica van Gieson staining, the elastic lamina was confirmed to have no venous component. a) Hematoxylin and Eosin staining. b) Elastica van Gieson staining. b-1) low-magnification image: the black square area shows the high magnification image shown below; b-2) high-magnification image: the yellow arrows show the elastic lamina.

vated lesion was formed due to thrombosis near the Dieulafoy's lesion (5). Endoscopically, type 2 lesions in the Yano-Yamamoto classification may progress to an SMT-like lesion due to thrombosis and fibrosis. Onogi et al. reported a case of arteriovenous malformation that formed an SMT-like finding accompanied by a large arterial vessel with inner thrombosis (6). In the present case, the blood vessel was considered to have been dilated because of arterial thrombosis of unknown etiology, creating an SMT-like lesion endoscopically.

Although small intestinal vascular lesions have been classified into three pathological patterns (angioectasia, Dieulafoy's lesion, and arteriovenous malformation), this case might be one of Dieulafoy's lesion, as there was no venous component. There have been several case reports of submucosal aneurysm of the small intestine, and the pathologic findings might be the same as in the present case (7-9). The formation of a hemispherical submucosal tumor-like lesion endoscopically may have been due to the thrombosis in the arterial vessel causing focal arterial dilation. Generally, it has been reported that Dieulafoy's lesion often appeared endoscopically as a type 2 lesion in the Yano-Yamamoto classification (1). Because a hemispherical SMT-like lesion derived from Dieulafoy's lesion was seen in the present case as well as in others reported in the literature, the endoscopic classification of small intestinal vascular lesions might require reconsideration. Imamura et al. referred to such a lesion as "Dieulafoy's vascular malformation" based on a histopathological assessment (10). To establish a new classification of hemispherical SMT-like protrusions with an exposed vessel, further cases will need to be accumulated.

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

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