



Identifying Small Bowel Gastrointestinal Stromal Tumor as the Culprit Lesion in Obscure Gastrointestinal Bleeding: Emphasis on Angiographic Findings

원인불명의 위장관 출혈을 보인 소장 위장관 기질종양 식별: 혈관조영술 소견의 강조

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Gastrointestinal stromal tumors (GISTs) are not uncommon and often cause gastrointestinal bleeding. GISTs occurring in the small intestine are occasionally difficult to identify by endoscopy and CT. In this case, the patient underwent CT three times before surgery, and the lesion was found to be located in a different area of the abdominal cavity on each CT scan. Moreover, the lesion was missed in the first two CT images because it was difficult to distinguish it from the nearby collapsed small intestine. The lesion was eventually detected through angiography; however, the correct diagnosis and treatment were delayed for 3 years because it was mistaken for a vascular malformation, which is the most common cause of obscure GI bleeding in elderly patients. This report emphasizes the need for interventional radiologists to be updated and vigilant of the angiographic features of GISTs to make an accurate diagnosis and establish a management strategy.

Index terms Gastrointestinal Stromal Tumor; Small Intestine; Gastrointestinal Hemorrhage; Angiography

INTRODUCTION

Gastrointestinal stromal tumors (GISTs) are not uncommon diseases that often cause

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gastrointestinal bleeding. When they occur in the small intestine, they are sometimes difficult to be identified by endoscopy and CT evaluation (1-4). The main source of obscure GI bleeding is angiodysplasia in elderly patients (5). Neoplasms also can be a cause of bleeding, especially GISTs, which have hemorrhagic potential due to ulceration of overlying mucosa (1, 2, 5). In general, when symptoms of GI bleeding appear, the tumor would have already reached a relatively large size or have shown distinct necrosis or ulceration that can be detected by CT (1). However, in this case, the lesion was not spotted by CT before angiography, because the lesion was difficult to distinguish it from the nearby collapsed small intestine and was not fixed in a certain area of the abdominal cavity. Although the existing literature has described the angiographic findings of small bowel GIST, most tumors were large enough to be easily found in CT, and the image quality of angiography was not satisfactory.

Herein, authors present detailed findings with good quality images obtained from the conventional angiography and literature review to provide helpful information for an accurate diagnosis and localization of the small bowel GISTs presenting with obscure GI bleeding in the future.

CASE REPORT

A 65-year-old male patient was sent to our emergency department complaining of melena and hematochezia for 24 hours. His vital sign was stable, his initial hemoglobin level was 6.5 g/dL, and other laboratory tests, including clotting, were normal. The source of the GI bleeding was not identified anywhere in the GI tract even after a comprehensive endoscopic evaluation including esophagogastroduodenoscopy, ileocolonoscopy and video capsule endoscopy.

Initial CT showed a lobular mass with relatively homogenous enhancement in the left upper abdominal cavity, measuring about 3.1 cm in the maximal diameter (Fig. 1A). Five days later, the mass was located in the left lower abdominal cavity on the follow-up CT. But the lesion was neither detected nor reported in the radiological report due to overlapping loops of nearby collapsed small bowel from both CT images.

The patient was referred to the interventional radiology department for the angiographic diagnosis and management due to refractory GI bleeding and persistent hemoglobin changes. Arterial phase of superior mesenteric artery angiogram showed a vascular tangle at the distal branches of the jejunal artery in the left lower abdomen. A well-circumscribed mass-like lesion with early draining veins was seen in delayed arterial phase. Numerous coiled and twisted feeding arteries were seen within the lesion. There were no contrast leaks, pseudoaneurysms or aneurysmal dilatation to be seen (Fig. 1B-D).

The lesion was misinterpreted as a vascular malformation because previous CT and endoscopy did not suspect any potential tumor development. A selective transarterial embolization was performed via feeders with polyvinyl alcohol particles (Contour 355–500 μ m, Boston Scientific, Natick, MA, USA) and three microcoils (Tornado 3 mm–2 cm; Cook Medical, Bloomington, IN, USA). As a result, the GI bleeding stopped and the hemoglobin levels were stabilized. The patient's postprocedural course was uneventful and he was discharged home on day 14 without any further surgical treatment.

Three years later, the patient revisited our emergency room with a complaint of hemato-

Fig. 1. Small bowel gastrointestinal tumor in a 65-year-old male manifesting as obscure gastrointestinal bleeding.

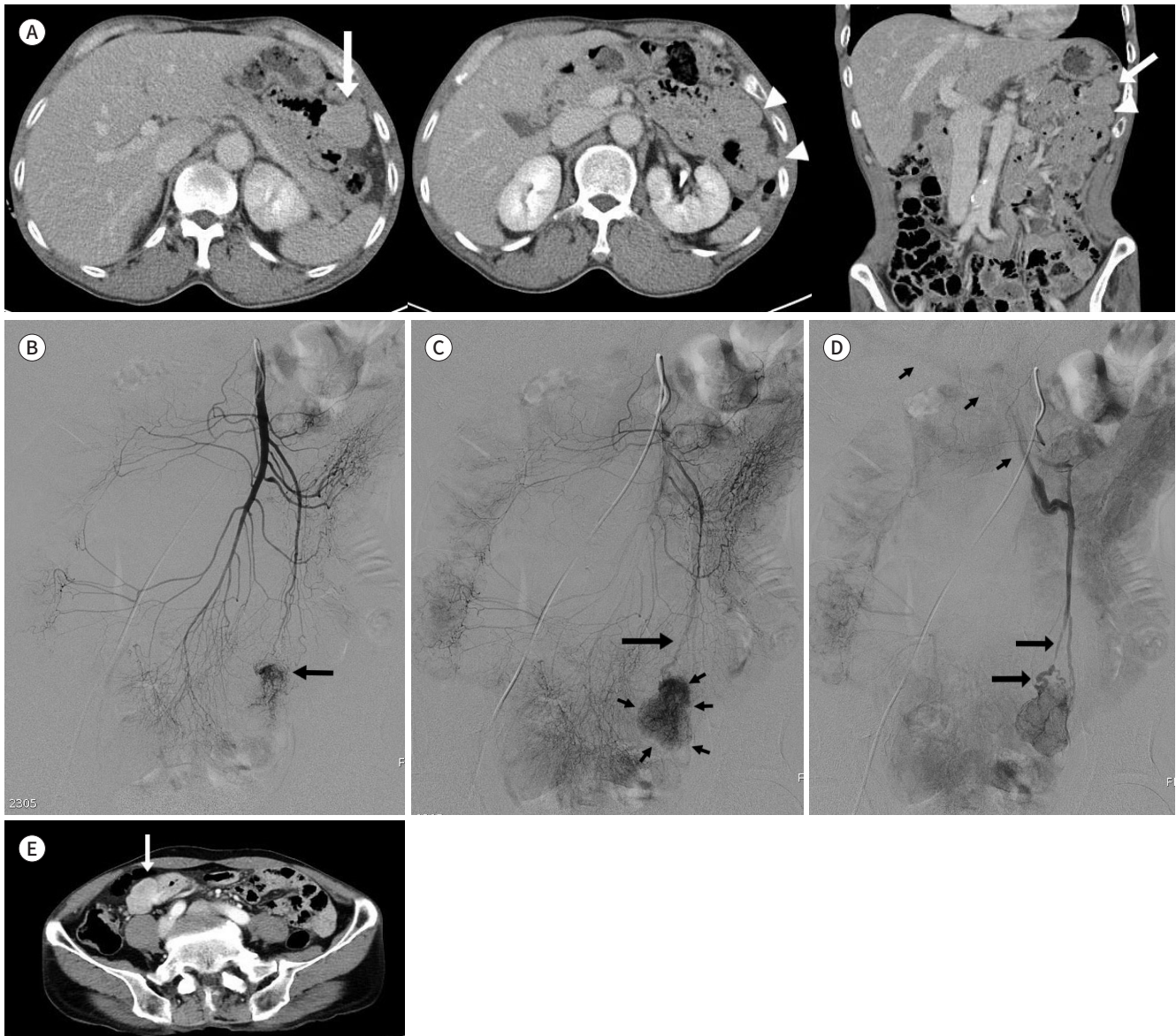
A. Initial CT scans reveal a lobular, homogeneous soft tissue mass (arrows) that could not be distinguished from the adjacent collapsed small intestine (arrowheads) in the left upper abdominal cavity.

B. The arterial phase of digital subtraction angiography reveals a vascular tangle (arrow) arising from the jejunal branches of the superior mesenteric artery.

C. Delayed arterial phase image reveals a well-circumscribed, lobular mass-like lesion (short arrows) with an early draining vein (long arrow). Abundant twisted, coiled feeding arteries are seen within the tumor.

D. In the venous phase, the markedly dilated, engorged vein (long arrows) appears to drain blood from the tumor to the portal system (short arrows).

E. CT image acquired three years later reveals an exophytic mass (arrow) arising from the small bowel loop in the right lower abdominal cavity.



chezia. CT image showed a lobular, enhancing mass in the right lower abdominal cavity (Fig. 1E). The patient underwent an exploratory laparotomy. An exophytic mass was found at jejunum, 30 cm distal to the Treitz ligament. Small bowel resection was performed. The specimen revealed a 3.8 cm × 3.0 cm × 2.7 cm sized, subepithelial mass protruding the antimesenteric border of small bowel with mucosal ulceration. Histology confirmed to have feature

of GIST, low risk with minimal pleomorphism (mitosis, < 5 per 50 high-power fields). At immunohistochemistry the tumor was positive for CD117 antigen (Fig. 1F).

This case report was approved by the local Institutional Review Board, and the requirement for written informed consent was waived (IRB No. 2021-06-008).

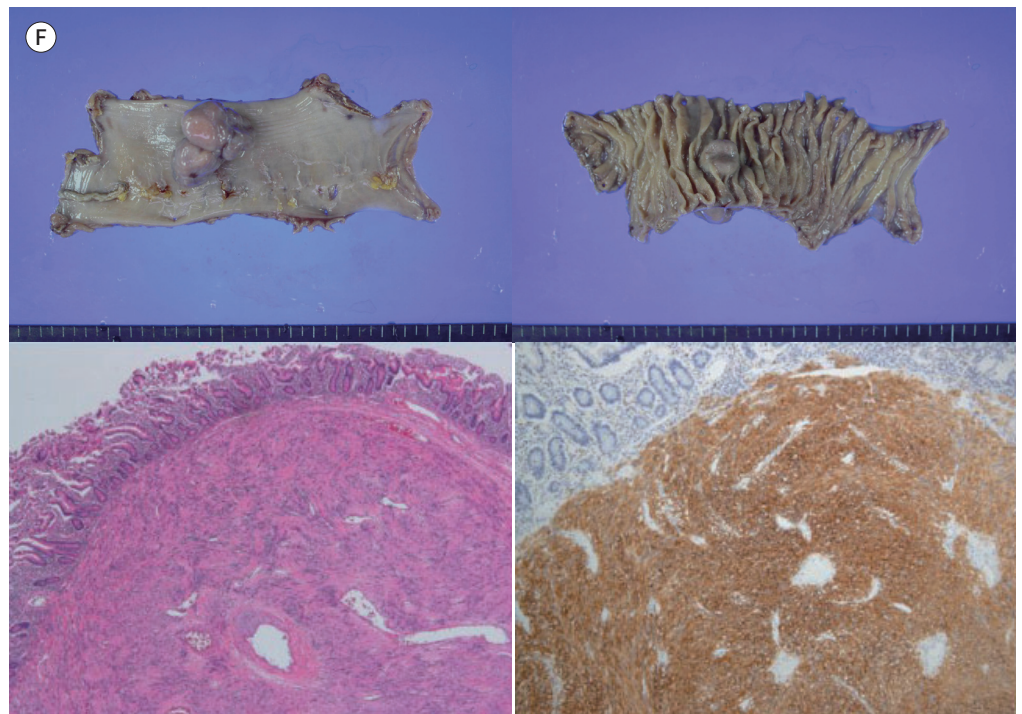
DISCUSSION

The small bowel is the second most common site (20%–35%) in GIST that develops in the GI tract after the stomach (40%–60%) (1, 2). Diagnosis of small bowel GISTs is sometimes delayed because of its relatively low incidence, non-specific symptoms and signs, and relative inaccessibility of the small bowel compared to the conventional endoscopy (2).

It is also difficult to detect a small bowel lesion using only CT because if a small bowel GIST is small in size and is not accompanied by pronounced necrosis, it can be confused with surrounding collapsed intestine in the CT images. In addition, since the jejunum and ileum are not fixed, and move in the abdominal cavity by being suspended by the mesentery; diagnosis may be more difficult if the lesion is moved from the last CT image, as it was in this case. According to previous reports, the rate of a provisional diagnosis of a small bowel GIST through CT was 54.8% (2) and the detection rate of a CT for a primary small bowel GIST was 67% (3).

Fig. 1. Small bowel gastrointestinal tumor in a 65-year-old male manifesting as obscure gastrointestinal bleeding.

F. Pathologic features. On gross examination, the specimen appears to be a subepithelial mass protruding into the antimesenteric border of the small bowel with mucosal ulceration. Hematoxylin and eosin staining (left, $\times 40$) shows spindle-shaped cells in the small bowel submucosa and wall. Few mitotic figures are seen (< 5 per 50 high-power fields). Immunohistochemical staining (right, $\times 40$) shows CD 117 (c-KIT)-positive tumor cells.



In this case, the patient underwent CT three times before surgery and from each CT image the lesion was located in a different area of abdominal cavity and was missed in the first two CT scans because it was difficult to distinguish it from the nearby collapsed small intestine.

GISTs have a hemorrhagic potential and GI bleeding is a relatively common presentation in patients with GISTs (1). The most common clinical manifestation of a symptomatic small bowel GIST is the obscure GI bleeding (2). Compared to other GI tract tumors, GISTs are relatively fragile and have rich blood vessels. As the tumor grows, it can cause mucosa and submucosa destruction as well. Digestive juices and fecal transmission happen together in combination to these histologic characteristics, which will result in ulceration in the overlying mucosa of GIST, and it will eventually lead to intraluminal bleeding (1).

Transarterial angiography can be useful when the endoscopy and conventional imaging fails to detect and localize the small bowel GISTs with intraluminal bleeding (4). It shows a coiled or hair-like appearance in the abundant, numerous feeding arteries without obvious hypertrophy. The margin of the tumor is well-circumscribed and enlarged draining veins develop early and merge into the portal system (4, 6). Some GIST with high risk or a large necrotic portion can show partially indistinct margin (6). Contrast leakage, pseudoaneurysm or aneurysmal dilatation is rare (4). At first glance, these findings are similar to those of angiodysplasia or arteriovenous malformation (note, terms such as vascular malformation, angiectasia, angiodysplasia, or arteriovenous malformation tend to be used interchangeably in the GI tract area while the aforementioned terms are used separately in other areas) which is the most common cause of obscure GI bleeding in elderly patients (5) and may require differentiation. In the case of angiodysplasia or arteriovenous malformation, hypertrophy of the feeding arteries is more evident, and the margin of the lesion is unclear. If a tumor is suspected on angiography, it is necessary to review CT images.

Meanwhile, transarterial embolization could be a good preoperative option as a prompt hemostasis in GISTs with overt GI bleeding (7). In this case, GI bleeding immediately stopped without complications by embolization. But, it is clear that embolization cannot be the curative treatment considering that there was no change in the size of the tumor and the GI bleeding recurred after 3 years.

The poor prognostic factors of GISTs are known to be a large tumor size, high mitotic count, non-gastric location, male sex, tumor rupture, and tumor bleeding (1, 8). Virtually GIST has malignant potential regardless of its size (8). Therefore, early detection and correct treatment of GIST is very important to the patient's prognosis.

When evaluating obscure GI bleeding, it is important to keep in mind the possibility of a small bowel GISTs even if it was not detected by endoscopy and CT. Familiarity with the angiographic findings of GISTs can help making an appropriate interpretation of the lesion and planning treatment strategies for patients with GISTs.

Author Contributions

Conceptualization, C.M.J.; investigation, C.H.I., C.M.J.; project administration, C.H.I., C.M.J.; resources, C.M.J., K.B.M., N.H., C.S.K.; supervision, C.M.J.; visualization, C.H.I., C.M.J., K.B.M.; writing—original draft, C.H.I., C.M.J.; and writing—review & editing, C.M.J., K.B.M., N.H., C.S.K.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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REFERENCES

1. Liu Q, Kong F, Zhou J, Dong M, Dong Q. Management of hemorrhage in gastrointestinal stromal tumors: a review. *Cancer Manag Res* 2018;10:735-743
2. Zhou L, Liao Y, Wu J, Yang J, Zhang H, Wang X, et al. Small bowel gastrointestinal stromal tumor: a retrospective study of 32 cases at a single center and review of the literature. *Ther Clin Risk Manag* 2018;14:1467-1481
3. Nakatani M, Fujiwara Y, Nagami Y, Sugimori S, Kameda N, Machida H, et al. The usefulness of double-balloon enteroscopy in gastrointestinal stromal tumors of the small bowel with obscure gastrointestinal bleeding. *Intern Med* 2012;51:2675-2682
4. Chen YT, Sun HL, Luo JH, Ni JY, Chen D, Jiang XY, et al. Interventional digital subtraction angiography for small bowel gastrointestinal stromal tumors with bleeding. *World J Gastroenterol* 2014;20:17955-17961
5. Gunjan D, Sharma V, Rana SS, Bhasin DK. Small bowel bleeding: a comprehensive review. *Gastroenterol Rep (Oxf)* 2014;2:262-275
6. Fang SH, Dong DJ, Zhang SZ, Jin M. Angiographic findings of gastrointestinal stromal tumor. *World J Gastroenterol* 2004;10:2905-2907
7. Koo HJ, Shin JH, Shin S, Yoon HK, Ko GY, Gwon DI. Efficacy and clinical outcomes of transcatheter arterial embolization for gastrointestinal bleeding from gastrointestinal stromal tumor. *J Vasc Interv Radiol* 2015;26:1297-1304.e1
8. Tirumani SH, Baheti AD, Tirumani H, O'Neill A, Jagannathan JP. Update on gastrointestinal stromal tumors for radiologists. *Korean J Radiol* 2017;18:84-93

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위장관기질종양(gastrointestinal tumor; 이하 GIST)은 종종 위장관 출혈의 원인이 되는 드물지 않은 질환이지만 소장에 생기는 경우 내시경 및 전산화단층촬영(이하 CT)에서 발견이 어려울 수 있다. 본 증례에서 환자는 수술 전 총 3회의 CT를 시행 받았으나, 병변은 매번 복강의 다른 부위에 위치해 있었고, 허탈된 소장과 구분이 어려워 처음 2번의 CT에서는 발견하지 못했다. 혈관조영술에서 병변이 보였지만, 고령의 환자에서 원인불명 위장관 출혈의 가장 흔한 원인인 혈관기형으로 잘못 해석되어 올바른 진단과 치료가 3년간 지연되었다. 본 증례를 통해, GIST의 정확한 진단 및 치료 전략 수립을 위해 혈관조영술 소견을 강조하고자 한다.

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