

# Spontaneous peeled ileal giant lipoma caused by lower gastrointestinal bleeding

## A case report

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### Abstract

**Rationale:** Gastrointestinal subepithelial tumors (SETs) with endoscopic features such as ulceration, a red color change, a peeled mucosal layer, and spontaneous bleeding could have malignant potential. However, we encountered a case of a lipoma that presented features different from the generally known features of gastrointestinal SETs. Therefore, we report an interesting rare case of a terminal ileal giant lipoma with a unique feature of spontaneous peeled ulceration on the surface on endoscopy that caused gastrointestinal bleeding.

**Patient:** An 82-year-old woman with a 1-week history of abdominal pain and hematochezia presented to our hospital.

**Diagnoses:** Ileocolonoscopy revealed a SET with a peeled surface and erythematous and ulcerative mucosal changes as well as exposed a submucosal mass at the terminal ileum. Macroscopically, the lesion appeared as a yellowish pedunculated polypoid mass measuring 3 × 2 cm with a peeled mucosal ulceration. Histopathological findings revealed a submucosal lipoma of the terminal ileum.

**Intervention:** We thought that the endoscopic finding indicated malignant SETs or those with malignant potential rather than benign SETs. Therefore, the patient underwent an elective laparoscopic ileocecectomy.

**Lessons:** We encountered a lipoma that did not present with the typical features of gastrointestinal SETs. Our findings suggest that clinicians should consider that benign SETs in the terminal ileum may present with various endoscopic findings similar to those of malignant SETs, which can cause fatal symptoms and signs.

**Abbreviations:** APCT = abdominopelvic computed tomography, CT = computed tomography, EGD = esophagogastroduodenoscopy, FDG = F-fluoro-2-deoxy-D-glucose, GIST = gastrointestinal stromal tumor, PET = positron emission tomography, SETs = subepithelial tumors.

**Keywords:** gastrointestinal bleeding, lipoma, small intestine, subepithelial lesions, terminal ileum

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## 1. Introduction

Gastrointestinal subepithelial tumors (SETs) may present as either benign or malignant. Therefore, it is important to distinguish benign lesions from those that are malignant. The first step of differential diagnosis is a meticulous endoscopic evaluation when SETs are found in the endoscopy. However, the endoscopic features of a SET in terminal ileum have been rarely reported, except in case reports. The reason for this is that, although the whole small intestine including the terminal ileum occupies >90% of the surface of the gastrointestinal (GI) tract, only 3% to 6% of all GI tumors occur in the small intestinal lumen, and SETs of terminal ileum are rare.<sup>[1,2]</sup> They may be found incidentally, but it may also manifest itself as various symptoms and signs. Vague and nonspecific symptoms include vomiting, nausea, and pain related to small bowel obstruction. Intussusception, perforation, or bleeding may also occur.<sup>[3–6]</sup>

Subepithelial lipomas account for 4% of the 4000 cases of benign GI neoplasm, and 64% of GI lipomas reportedly occur in the large intestine, 31% in the small intestine, 3% in the stomach, and 12% in the esophagus.<sup>[7,8]</sup> They are found mainly in the ileum and duodenum of the small intestine, but their prevalence in the terminal ileum remains unclear. In addition, the intestinal lumen is difficult to access using conventional esophagogastroduodenoscopy (EGD) and colonoscopy.

Recently, video capsule endoscopy and double balloon enteroscopy have been introduced,<sup>[1,9–12]</sup> but these techniques

**Table 1****Complete blood count and coagulation test.**

	On admission	Normal range
Hemoglobin, g/dL	6.7	11.5–15.5
Hematocrit %	20.8	34.5–46.5
Red blood cells, $\times 10^{12}/L$	2.19	3.8–5.0
Mean corpuscular volume, fL	95	82–98
Mean corpuscular hemoglobin, pg	30.6	27–33
Mean corpuscular hemoglobin concentration, g/dL	32.2	32–36
Red cell distribution width %	14.9	11.5–14.5
White blood cells, $\times 10^9/L$	9.33	3.7–9.5
Platelets, $\times 10^9/L$	287	150–400
Mean platelet volume, fL	9.7	9.3–13.0
Platelet distribution width %	10.5	11.6–13.7
Prothrombin time, INR	0.98	0.88–1.19
Partial thromboplastin time, second	26.5	27–39.5

are only able to be used in some patients because the equipment is expensive. Therefore, endoscopic characteristics of SETs in the terminal ileum are rarely reported except in case reports.<sup>[13,14]</sup> For this reason, little is known about the endoscopic appearance of SETs in the terminal ileum. Therefore, we apply the endoscopic characteristics of SETs reported in the stomach, esophagus, and duodenum in most cases.

Benign GI SETs are generally covered with the same mucosa as the surrounding tissue endoscopically.<sup>[15]</sup> GI SETs with endoscopic features such as ulceration, red color change, pilled mucosal layer, and spontaneous bleeding could have malignant potency.<sup>[16,17]</sup> However, we recently experienced a case of benign SETs in the terminal ileum that we considered malignant tumors because of the interesting endoscopic findings. Therefore, here, we report a case of an ileal giant lipoma with a spontaneous peeled mucosal surface that caused lower GI bleeding in an 82-year-old Korean woman.

## 2. Case report

A 82-year-old woman was referred to our gastroenterology clinic on September 22, 2016 due to a 1-week history of abdominal pain and hematochezia. She had no medical history, other than hypertension and visual disturbance due to macular degeneration, and she had no family cancer history. She

underwent EGD at a private clinic. The findings of EGD showed mild erosive gastritis, but no cause of the hematochezia was found.

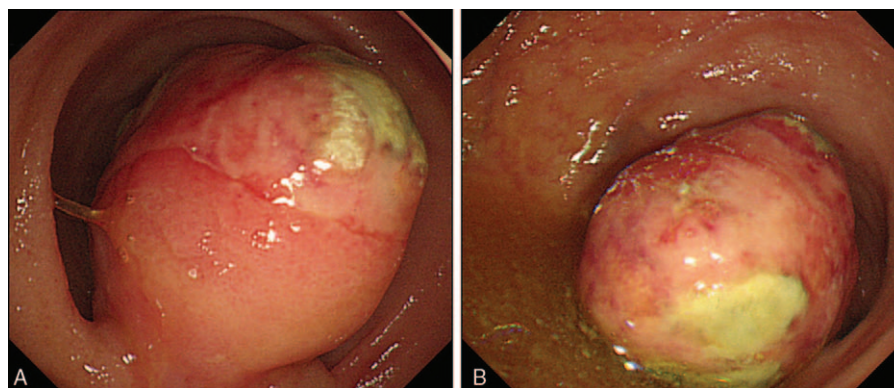
Her physical examination showed a pale conjunctiva and mild tenderness of the right lower abdomen. She had no chills or fever. Her blood pressure was 99/57 mm Hg, pulse rate was 88/min, and body temperature was 36.5°C. She had clear consciousness, but exhibited an acute ill appearance.

A complete blood count on the day of admission revealed the following: hemoglobin, 6.7 g/dL; hematocrit, 20.8%; white blood cell count,  $9.33 \times 10^9/L$ ; and platelet count,  $287 \times 10^9/L$  (Table 1). The patient's prothrombin time and partial thromboplastin time/international normalized ratio were normal. The results of peripheral blood smear excluded iron deficiency anemia and anemia of chronic disease. The blood chemistry results were as follows: serum total protein, 5.9 g/dL (normal, 6–9.2 g/dL); albumin, 3.3 g/dL (normal, 3.5–5.2 g/dL); aspartate aminotransferase, 19 U/L (normal, 10–40 U/L); alanine transaminase, 11 U/L (normal, 5–40 U/L); blood urea nitrogen, 26 mg/dL (normal, 8–22 mg/dL); creatinine, 0.7 mg/dL (normal, 0.5–1.2 mg/dL); and C-reactive protein, 0.16 mg/dL (normal, 0–0.5 mg/dL).

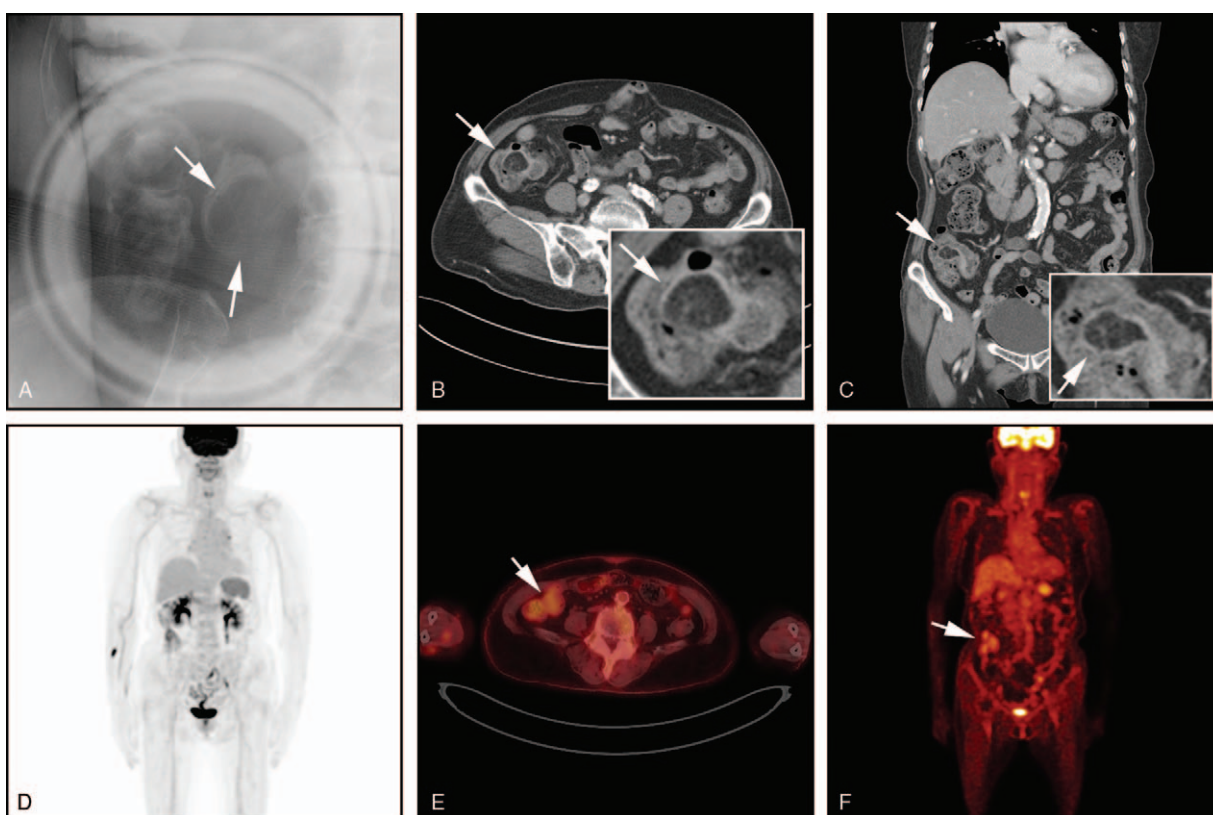
An ileocolonoscopy revealed, regarding the SET, that the surface was peeled with erythematous and ulcerative mucosal changes and a submucosal mass exposed at the terminal ileum (Fig. 1A and B). Although we attempted to perform a biopsy to ensure an accurate diagnosis, only a tiny amount of tissue was retrieved because the mass was very movable rather than fixed.

On small bowel gastrografin study, the area of concern presented as radiolucent, intramural lesions with compression, with no evidence of obstruction (Fig. 2A). The abdominopelvic computed tomography (APCT) scan showed well-demarcated 30-mm tumor in the terminal ileum (Fig. 2B and C). Positron emission tomography (PET)-CT revealed a slightly increased 18-F-fluoro-2-deoxy-D-glucose (FDG) uptake in the terminal ileum ( $mSUV=2.59$ ) (Fig. 2D–F).

At this point, the patient underwent an elective laparoscopic ileocecectomy because the SET of the terminal ileum was considered to be the lower GI bleeding source, and had a possibility that it could grow or have malignant potential. In macroscopic findings, the lesion appeared pedunculated polypoid mass with peeled mucosal ulceration (Fig. 3A and B). The histopathological findings revealed submucosal lipoma of the



**Figure 1.** Endoscopic view of the terminal ileal mass. (A) Side view of an endoscopy. (B) Top view of an endoscopy. (A, B) The ileocolonoscopy revealing that more than half of the surface of the terminal ileal mass was peeled, and that ulcerative mucosal changes had formed, and that the submucosal tissue was exposed at the terminal ileum.



**Figure 2.** Diagnostic imaging findings. (A) Small bowel study. Enterography using gastrografin showing a very radiolucent, well-circumscribed, intramural filling defect compressing the terminal ileum but no evidence of obstruction. (B, C) The contrast-enhanced portal venous CT image (transverse (B), coronal (C) image). The abdominopelvic CT scan showing a round, smooth, and well-demarcated 30-mm tumor with fat attenuation in the submucosal layer of the terminal ileum. These are well-circumscribed intraluminal masses with homogenous fat attenuation. (D–F) PET-CT image. PET revealed slightly increased 18-FDG uptake in the terminal ileum. CT = computed tomography, FDG = F-fluoro-2-deoxy-D-glucose, PET = positron emission tomography.

terminal ileum (Fig. 3C and D). She was discharged uneventfully on the eighth day after the operation.

### 3. Discussion

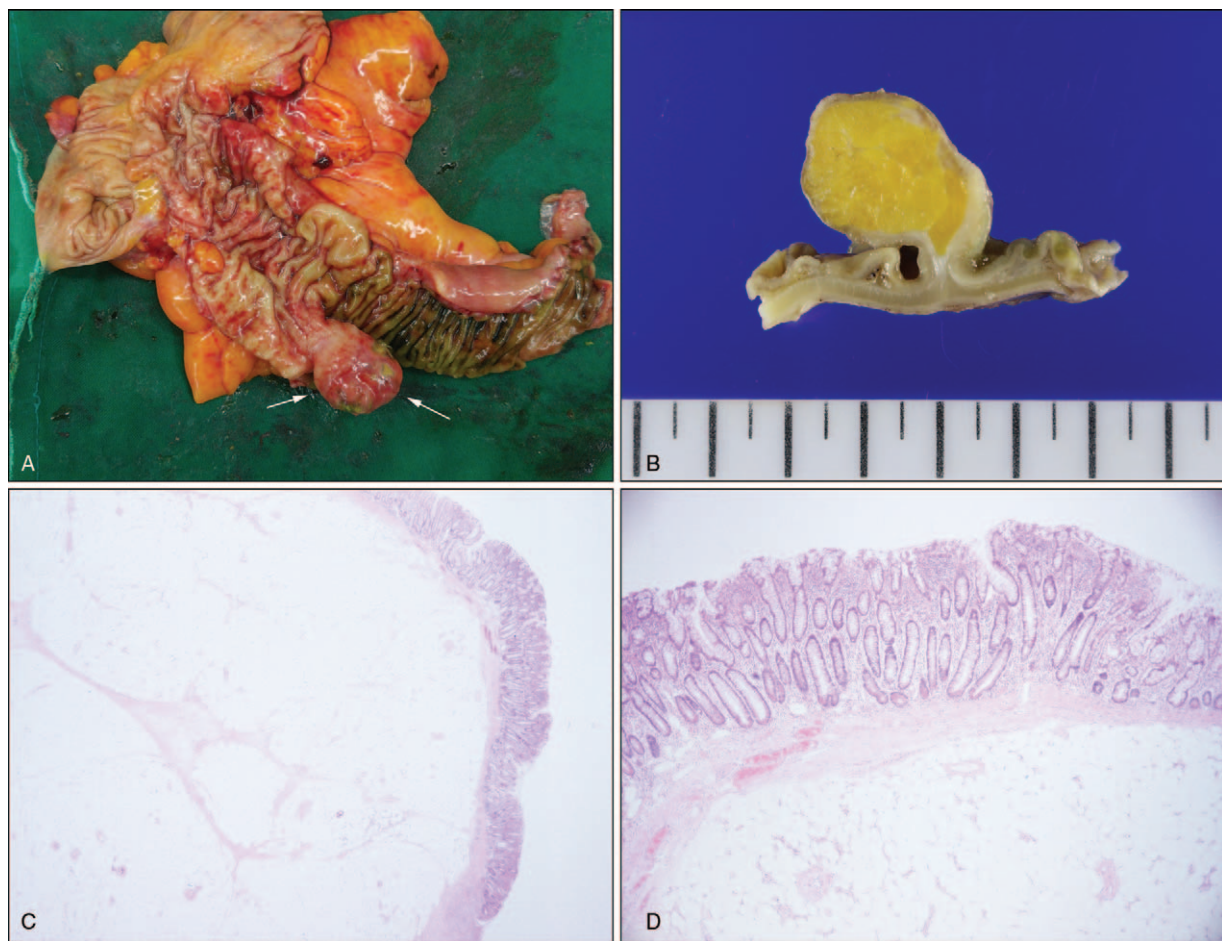
Although the most important feature of the SETs is that they are surrounded by normal mucosa, some SETs may have a change in the surrounding mucosa. More careful observation is needed when endoscopic findings are unusual. In particular, if SETs show a changed surface that is not covered by the intact normal mucosal or a rapid change in size, more aggressive examination and treatment are needed, regardless of size.<sup>[18,19]</sup> If there is an erosion or ulceration on the surface of hard SETs, we suspect more susceptible malignant SETs or those with malignant potential such as GI stromal tumor (GIST), neuroendocrine tumors, or cancer. There are some reports of endoscopic features of small bowel malignant SETs with surface change.<sup>[16,17]</sup>

GISTs, carcinoid tumors, and lymphoma are malignant, or have the potential to become malignant.<sup>[15–17]</sup> These tumors sometimes show submucosal masses with red color change, erosion, and ulceration endoscopically.<sup>[16,17]</sup> In the present case, ileocolonoscopy showed that the SET surface was peeled with erythematous and ulcerative mucosal changes and submucosal mass exposed at terminal ileum. Therefore, we thought that this endoscopic finding meant malignant SETs or those with malignant potential, rather than benign SETs.

In the present case, the final diagnosis was confirmed as lipoma. Generally, intestinal lipoma is seen as submucosal masses with round or hemispherical shapes, a smooth surface, well-demarcated margin, and/or encapsulated yellowish surface.<sup>[13,14,20]</sup> However, this case did not show this form, and instead showed an interesting endoscopic findings with more than one-half of the surface peeling off and ulceration. In addition, the cushion and pillow signs, which are mainly seen in lipoma,<sup>[21]</sup> were not clear and did not help endoscopic diagnosis.

Conventional endoscopic findings of the ileum located in the lipoma are rarely reported, but there are a few reports of this unique type.<sup>[13,14]</sup> These cases involve those of endoscopic removal of the lipoma with an intact mucosal surface at the terminal ileum. Unlike these cases, the mucosal surface of the terminal ileal mass in this case had peeled naturally and an ulcer had formed. We suggest that the ulcer caused by the surface being peeled off is thought to be damage of the surface caused by friction, because the size is large and the transit time of the small intestine is very fast. In addition, it is thought that this spontaneous ulcerative peeled surface caused GI bleeding.

Benign SETs are treated according to their clinical symptoms and size. If a patient with small bowel lipomas is asymptomatic, supportive treatment is generally recommended.<sup>[20]</sup> However, the mainstay treatment of patients with symptomatic SETs is surgery.<sup>[20]</sup> The endoscopic treatment of patients with symptomatic small bowel lipoma is an alternative if small bowel lipoma can be removed by endoscopic resection.<sup>[13,14,22]</sup> In the present



**Figure 3.** Histopathology of the terminal ileal tumor. (A) The macroscopic view of the resected specimen demonstrating a round reddish tumor with a peeled mucosal surface. (B) In the longitudinal section of the tumor, a large yellow pedunculated polypoid mass measuring 3 × 2 cm is seen that resembles lipoma. (C, D) Microscopic findings revealing the submucosal lipoma of small intestine. The tumor is microscopically composed of mature fat. Mature adipocytes with compact eccentric nuclei and abundant lipid-filled cytoplasm are evident in this lipoma [hematoxylin and eosin staining: ×4 (C) and ×40 (D)].

case, the patient underwent an elective laparoscopic ileocecectomy because the lipoma of the terminal ileum was considered as the lower GI bleeding source; there was a possibility of obstruction or intussusception because of large size; and there was a malignant potential in endoscopic findings. Histopathological findings revealed that the submucosal lipoma consisted of mature fat.

They commonly appear as an ovoid, delineated, and smooth mass in small bowel series.<sup>[23]</sup> They are described with a smooth, round, well-demarcated lesions with a coefficient of fat attenuation on CT scan.<sup>[23]</sup> In the present case, findings of APCT scan and small bowel series are compatible with these features. PET-CT revealed slightly increased 18-FDG uptake at solid portion, as shown in APCT.

It is known that lipomas lack FDG accumulation on PET scans.<sup>[24]</sup> However, there are a few reports in which PET uptake indicated a false-positive result.<sup>[24–26]</sup> We did not find any studies reporting on the uptake pattern of PET image in terminal ileal lipoma. We suggest that the 18-FDG uptake was slightly increased by the inflammatory reaction when the surface of the ileal lipoma was peeled off and the ulcer was formed.

The rate of terminal ileum intubation was routinely about 87% during colonoscopy.<sup>[27]</sup> If an intestinal bleeding is suspected, terminal ileum intubation was needed because some tumors of

the small intestine are located in the distal ileum. In the present case, we found a SET of the terminal ileum during colonoscopy through ileum intubation without enteroscopy.

#### 4. Conclusion

In conclusion, we have experienced a lipoma that looked different from generally known features of GI SETs. Therefore, we report an interesting rare case of terminal ileal giant lipoma, with a unique feature of spontaneous peeled ulceration on the surface on endoscopy that caused GI bleeding. It should be noted that benign SETs in the terminal ileum may also present with various endoscopic findings that look like malignancy, and can cause fatal symptoms and signs.

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