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Case Report

Hemorrhagic shock due to ruptured idiopathic intramural hematoma of the sigmoid colon—An autopsy case report ☆

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ABSTRACT

We describe herein an autopsy case involving fatal hemorrhagic shock due to idiopathic sigmoid colonic intramural hematoma rupturing into the abdominal cavity. Antemortem computed tomography revealed a mass lesion in the sigmoid colon and a large amount of hemoperitoneum. On forensic autopsy, intramural hematoma of the sigmoid colon with ruptured serosa was identified, while the mucosa remained intact. Microscopically, hematomas were apparent, mainly in the muscularis propria. We diagnosed the cause of death as hemorrhagic shock due to idiopathic intramural hematoma of the sigmoid colon rupturing into the abdominal cavity. Although the patient had been receiving peritoneal dialysis, no relationship was identified between dialysis catheters and sigmoid colon intramural hematoma rupture. On computed tomography, the mass lesion was initially considered a submucosal neoplastic lesion or endometriotic lesion. Intramural hematoma should be considered as a differential diagnosis.

Introduction

Intramural hematoma of the colon is rare, with more than 90% of gastrointestinal intramural hematomas occurring in the duodenum and only 5% involving the colon [1,2]. For example, esophageal [3–5], gastric [6], and duodenal [7] intramural hematomas have been reported in the *Radiology Case Reports*. Gastrointestinal intramural hematomas mainly develop in the submucosa or muscularis propria [6]. The most common cause of intramural hematoma of the colon is blunt abdominal trauma, followed by anticoagulant therapy and blood dyscrasia [8]. We present herein the case of a woman in whom idiopathic intramural hematoma of the sigmoid colon, mainly

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between the inner circular and outer longitudinal muscle layers, ruptured into the abdominal cavity, causing death by hemorrhagic shock. As the patient had been receiving continuous ambulatory peritoneal dialysis (CAPD), from a forensic perspective, we had to elucidate the relationship between dialysis catheters and intramural hematoma of the sigmoid colon.

Case report

Case history

The patient was a premenopausal woman in her 50s who had been diagnosed with diabetes 8 years earlier. She had been receiving CAPD for diabetic nephropathy at home for about a year. In addition to diabetic nephropathy, the primary-care physician had noted hypertension and hyperlipidemia, but no abnormalities suggestive of hematological malignancies or coagulation/fibrinolytic disorders were obvious from blood test results.

The night before this event, according to dialysis records, the patient started peritoneal dialysis as usual. The next morning, she was found breathing but unconscious. At that time, drainage fluid from the peritoneal dialysis showed reddish discoloration and the dialyzer had issued an alarm and displayed the alert "poor drainage." She was taken to the emergency room and given mechanical chest compressions, endotracheal intubation, and adrenaline injections, but died about 3 hours later.

Findings from antemortem computed tomography

Antemortem non–contrast-enhanced computed tomography (CT) was performed in the emergency room (Fig. 1). A large amount of hemoperitoneum was evident. In addition, a mass lesion 3 cm in diameter was identified in the sigmoid colon (Figs. 1A, B, and D). The mass lesion showed homogeneous high attenuation compared to muscle tissue and a smooth margin (Figs. 1G and H). These findings for the mass lesion were considered inconsistent with cancer, but whether the lesion represented a submucosal neoplasm such as gastrointestinal stromal tumor or lymphoma or a non-neoplastic lesion such as intestinal endometriosis could not be determined.

A dialysis catheter was observed to the left of the mass lesion, separated by a 0.8-cm thickness of adipose tissue (Figs. 1A and B). Soft tissue displaying heterogeneous attenuation was seen on the right side of the mass lesion, which appeared to be a soft blood clot or fibrin (Figs. 1A, B, and D). Rupture and hemorrhage on the right side of the mass lesion were considered because soft blood clots and fibrin were unevenly distributed on the right side of the mass lesion. The tip of the dialysis catheter was located deep in the pouch of Douglas (Fig. 1F). No soft clot was seen at the site where the dialysis catheter penetrated the abdominal wall (Figs. 1C and E). Encapsulating peritoneal sclerosis is a known complication of peritoneal dialysis, but was ruled out in this case.

Although hemorrhage from the sigmoid colon was strongly suspected, as described above, the patient was taken to the emergency room in a state of shock, and was unable to undergo other examinations, including contrast-enhanced CT and colonoscopy.

Autopsy findings

Autopsy was performed 3 days postmortem. External examination showed that the patient was 155 cm tall and weighed 73 kg. Livor mortis was weak, and the conjunctivae and oral mucosa were pale. No abnormalities were evident at the insertion site of the dialysis catheter. No injuries other than those caused by the mechanical compressions for cardiopulmonary resuscitation were observed. No evidence of blunt trauma to the abdomen was seen.

Internal examination showed 4290 mL of bloody ascites, including soft blood clots and fibrin (Figs. 2A and B). However, since this volume also contained peritoneal dialysate, the measured amount of bleeding was considered inaccurate. A dark-red mass lesion 3 cm in diameter was identified in the sigmoid colon. The serosa over the lesion was ruptured and was considered the origin of bloody ascites (Fig. 3). The site of mass lesion rupture was far from the catheter. Further, the tip of the dialysis catheter was located deep in the pouch of Douglas. The cut surface of the sigmoid colon mass showed hematoma, and no lesions such as neoplasms were detected macroscopically. The mucosa overlying the lesion remained intact. Hemoglobin concentration was 3.1 g/dL and hematocrit was 11.4%. The patient displayed renal anemia antemortem, with a hemoglobin concentration of 7.6 g/dL and hematocrit of 20.8% at 3 weeks before her death. Blood volumes in various organs were low, and slight hemorrhage was observed in the left ventricular endocardium. These findings were considered to reflect lethal hemorrhage.

In addition, renal atrophy was observed and was considered consistent with diabetic nephropathy. Except for ruptured intramural hematoma of the sigmoid colon and bloody ascites, no lesions considered capable of causing death were obvious. Poisoning was also ruled out. CT and microscopy showed no findings suggestive of dialysis amyloidosis in the joints, spine, or other organs.

Microscopically, the mucosa of the sigmoid colonic lesion remained intact. Hematomas were localized from the submucosa to the subserosa, mainly between the inner circular and outer longitudinal muscle layers (Fig. 4). No pathology responsible for bleeding, such as neoplasm, amyloidosis, or angiodysplasia, was identified. Infiltration of inflammatory cells and fibrin (Figs. 2C and D) were confirmed, and the hematoma was considered fresh. No chronic inflammation suggestive of chronic catheter-related stimulation was found in the serosa of the sigmoid colon. Other microscopic findings included hyaline-like changes in efferent and afferent arterioles and fibrin caps in the renal glomeruli. Disrupted glomeruli were also seen. These findings were considered consistent with diabetic nephropathy.

From the above macro- and microscopic findings, we diagnosed the cause of death as hemorrhagic shock due to intramural hematoma of the sigmoid colon that had ruptured into the abdominal cavity. As for the cause of the intramural hematoma, no blunt trauma to the abdomen or underlying disease state was identified, so the case had to be considered

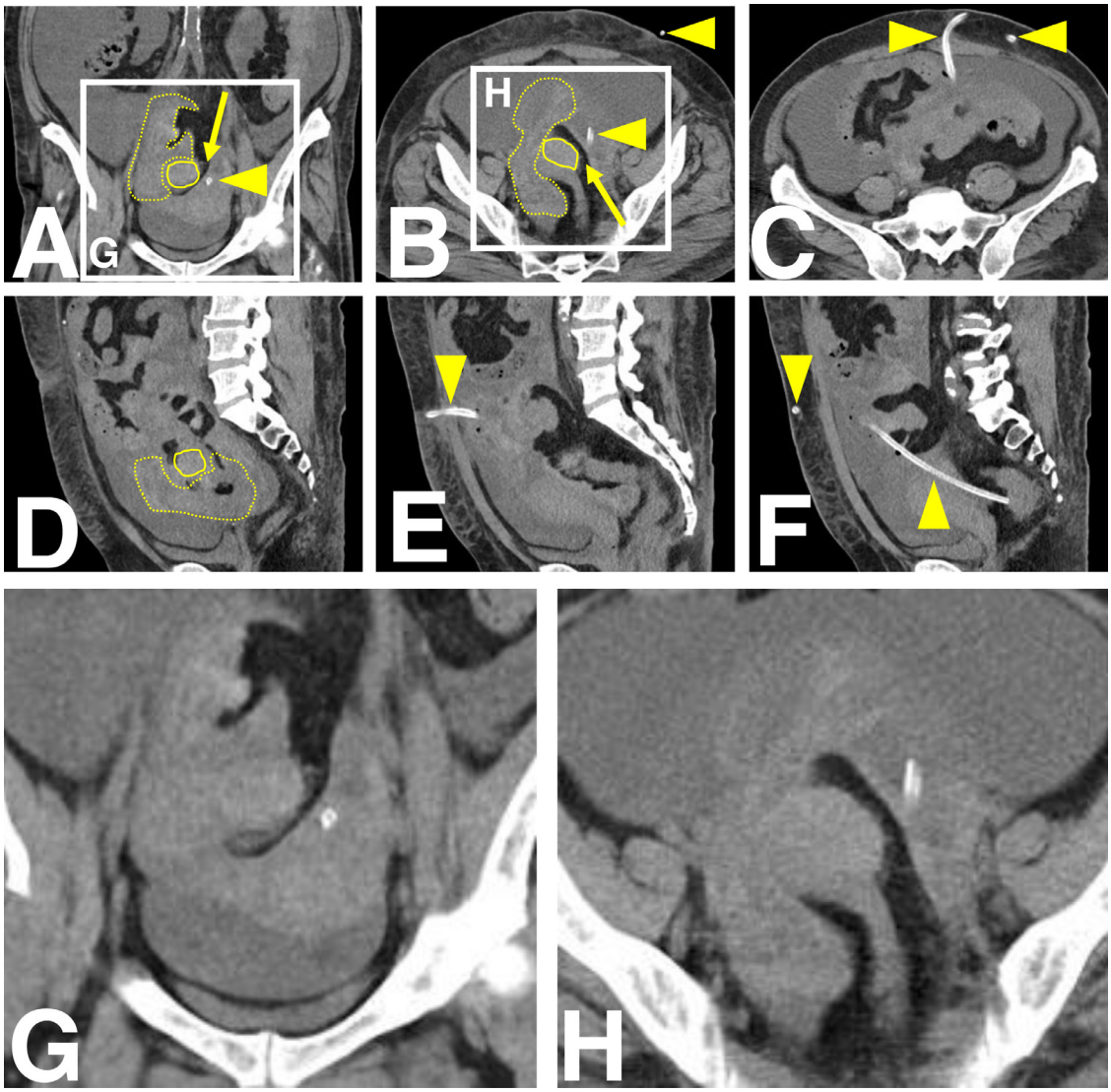

Fig. 1 – Antemortem non–contrast-enhanced CT images. Circles indicate the mass lesion in the sigmoid colon. Arrowheads indicate the dialysis catheter. Arrows indicate adipose tissue interposed between the mass lesion and dialysis catheter. Dashed circles indicate soft blood clots and fibrin. (A) Coronal-view reconstruction of abdominal computed tomography (CT). (B) Axial-view image from pelvic CT. (C) Axial image taken 4.5 cm cranial from Image B. (D) Sagittal-view reconstruction of abdominal CT. (E) Sagittal-view image taken 2.1 cm to the left of Image D. (F) Sagittal-view image taken 1.2 cm to the left of Image E. (G) Enlargement of Image A. (H) Enlargement of Image B.

idiopathic. No other lesions that could have caused death were evident.

Discussion

Intramural hematoma of the colon is rare [1], and clinical case reports have been described only sporadically. We were able to identify 28 clinical case reports in searches of PubMed and Scopus using the keywords "intramural hematoma" and "colon." Among these 28 cases, 1 patient died. One case involved postpolypectomy complications in which hemicolectomy was performed to include an intramural hematoma lesion, but the patient died 4 days after surgery [9]. In addition to these searches in the literature, 1 fatal case was reported in Japanese. The patient had a history of myelodysplastic syndrome and chronic heart failure, and was warfarinized. The lesion was resected after the patient developed an intramural hematoma of the colon, but he died 34 days after surgery due to heart and respiratory failure [10].

Etiology of intramural hematoma of the colon

The most common cause of intramural hematoma of the colon is blunt abdominal trauma, followed by anticoagulant therapy and blood dyscrasia [8]. In this case, the patient had received mechanical chest compressions but sigmoid colon

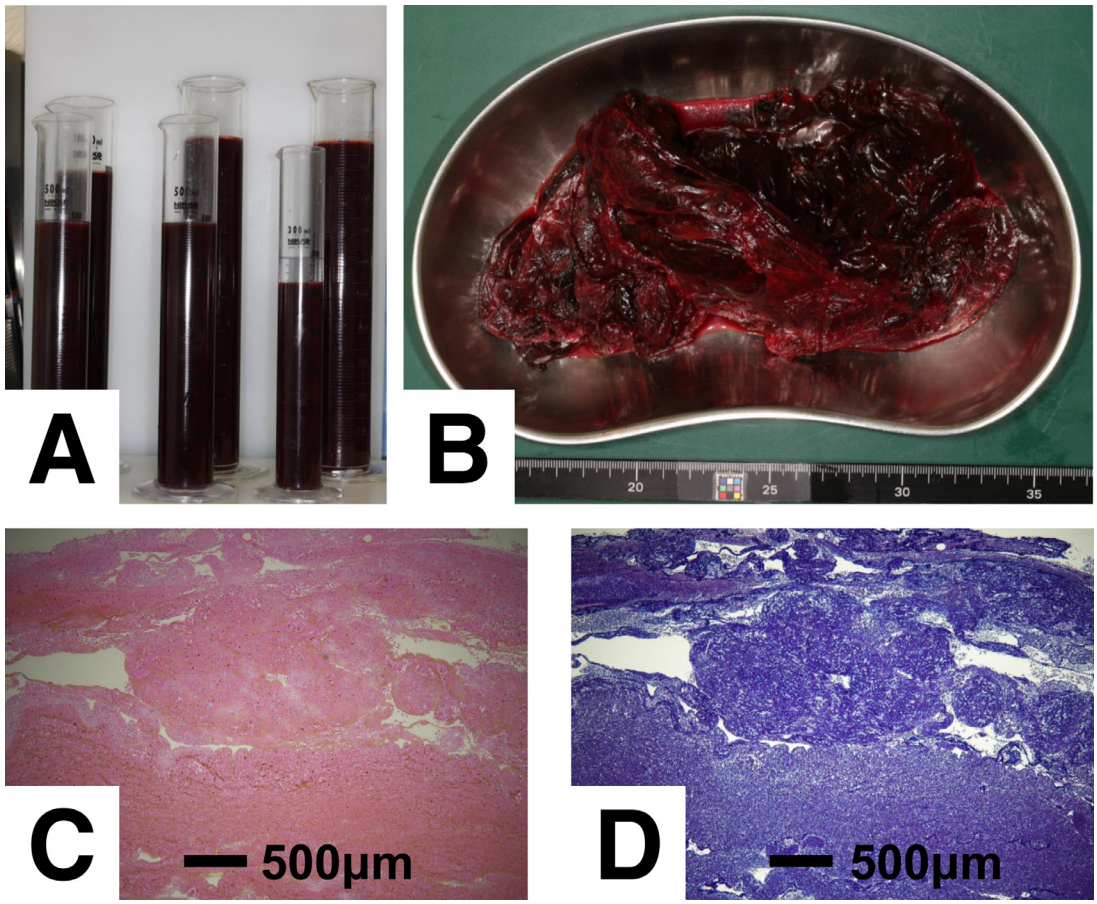

Fig. 2 – Bloody ascites, soft blood clots, and fibrin. (A) Bloody ascites (4290 mL). (B) Soft blood clots and fibrin. (C) Microscopic views of fibrin. Hematoxylin and eosin (HE), ×40. (D) Microscopic views of fibrin. Phosphotungstic acid hematoxylin, ×40.

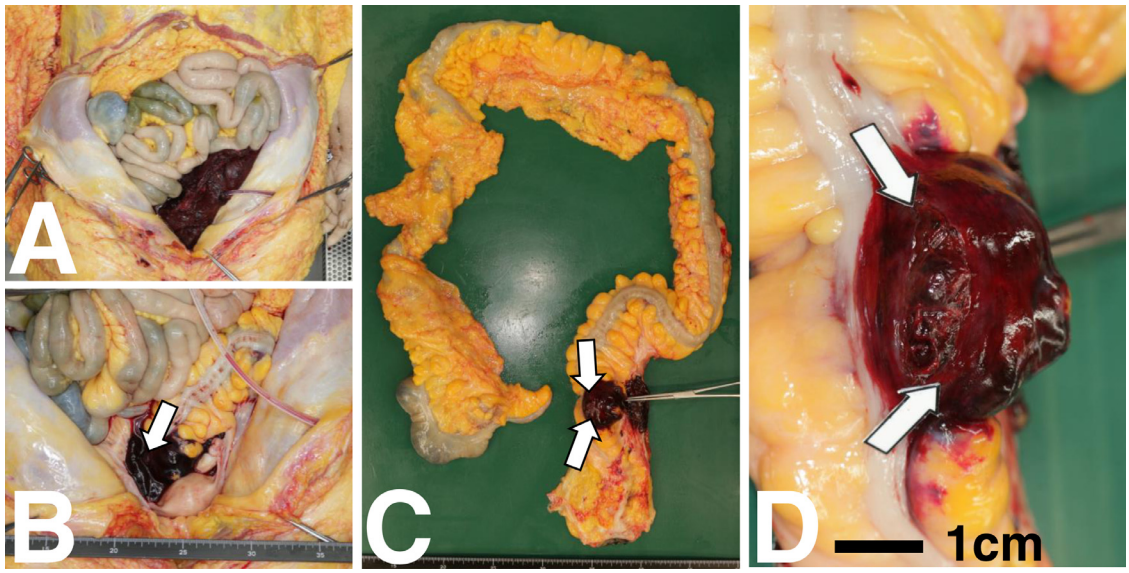

Fig. 3 – Arrows indicate the site of rupture of the sigmoid serosa. (A) Abdominal cavity. Most of the bloody ascites has been removed. (B) Most of the soft clot and fibrin removed from photo A. (C) Whole colon removed. (D) Close-up image of the rupture site in the sigmoid serosa.

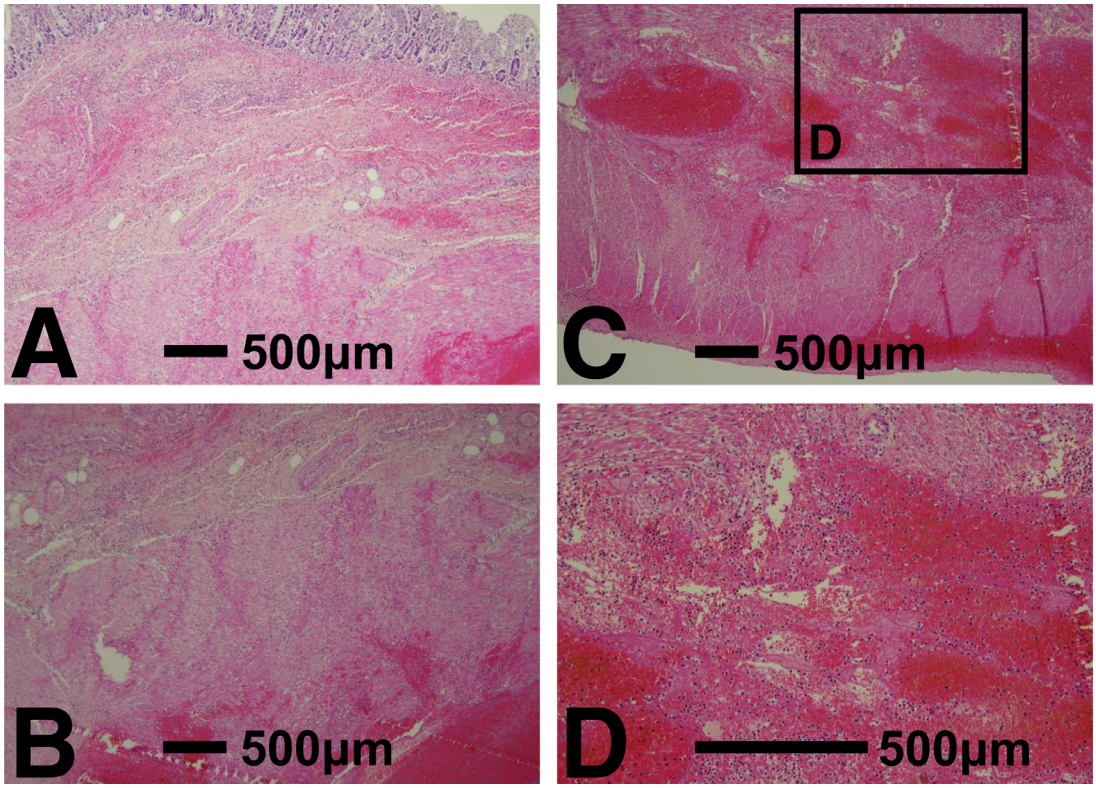

Fig. 4 – Microscopic views of intramural hematoma from the sigmoid colon. HE. (A) The mucosa is intact, with dense hematoma in the lower layer of the inner circular muscle seen in the lower right of the photograph. Slight bleeding is also observed in the submucosa. ×40. (B) Lower layer of Image A. Below the photograph, the serosa is ruptured. ×40. (C) An area in which hematoma is clearly visible between the inner circular and outer longitudinal muscle layers. Hematoma is also evident in the subserosa. The serosa is not ruptured in this area. ×40. (D) Enlargement of Image C. Inflammatory cells and fibrin are apparent. ×100.

injury was unlikely to have resulted as a complication of these [11]. Gastrointestinal hematomas are estimated to occur in 1 of every 250,000 anticoagulated patients [12]. The patient in our case did not show findings suggestive of blunt abdominal trauma, was not anticoagulated and had no obvious blood dyscrasias. Postpolypectomy status [9,13], endometriosis [14], and a background of acute cholecystitis [15] or intestinal pneumatosis [16] have been reported as other causes. In our case, those causes were all excluded by a comprehensive medical history and macro- and microscopic findings. We therefore had to classify this case as idiopathic, a cause representing about 1% of intramural hematomas [17].

Symptoms of intramural hematoma of the colon

The clinical symptoms associated with intramural hematoma of the colon are nonspecific, varying from tolerable abdominal pain to hemorrhagic shock [9]. Symptoms may differ depending on whether the bleeding remains intramural, ruptures and bleeds into the mucosa, or ruptures and bleeds outside the serosa. Cases of bleeding remaining intramural [8,18], cases of rupture and bleeding into the lumen resulting in hematochezia [19,20], and cases of rupture and bleeding into the abdominal cavity [21,22] have all been reported. Cases of intramural hematoma of the ascending colon bleeding in the retroperitoneum [23] and massive bleeding from intramural hematoma of the ascending colon extending to the intestinal lumen, abdominal cavity, mesentery, and retroperitoneum have also been reported [11].

Diagnosis and treatment of intramural hematoma of the colon

Nakamura et al. reported a case of idiopathic gastric intramural hematoma diagnosed using contrast-enhanced ultrasonography [6]. Because our patient was obese, sigmoid intramural hematoma may have been difficult to diagnose on ultrasonography.

Colonoscopy is useful for diagnosis [8]. Colonoscopy shows an elevated lesion covered with normal colonic mucosa and blue mucosa around the lesion, reflecting submucosal hematoma [17].

Hematoma is visualized as a high-attenuation mass compared to muscle tissue on CT in the acute phase, but the hematoma is gradually absorbed and attenuation thus decreases over time, requiring differentiation from other lesion such as cysts, abscesses, and necrotic tumors [21]. In our case, the mass showing high attenuation compared to muscle tissue detected on CT (Fig. 1) was considered consistent with acute hematoma. The patient also underwent dialysis 3

times a day, and drainage fluid from the previous dialysis was normal according to dialysis records. This time course and histopathological findings were also consistent with acute hematoma. We considered that contrast-enhanced CT contributes to the qualitative diagnosis of mass lesions and identification of bleeding sites.

Magnetic resonance imaging (MRI) findings of intramural hematoma have been reported [17,24]. On MRI, signal changes vary depending on the postbleeding interval [17,24]. The patient in this case was taken to the emergency room in a state of shock, with no time to perform MRI.

Intramural hematomas of the colon are usually found in the submucosal layer [6,10,11,17,25] and muscularis propria [6]. A case of idiopathic bleeding in the inner circular muscle layer has also been reported [21]. In this case, hematoma was mainly localized between the inner circular and outer longitudinal muscle layers. The cause of bleeding into the muscularis propria is unknown.

Surgical treatment is considered the gold standard by some [8,19], while conservative treatment remains the first choice according to others [10,21]. Of course, surgical treatment carries risks in patients on anticoagulant therapy or with underlying blood disorders [10].

Relationship between peritoneal dialysis and bleeding in this case

Takagi et al. [26] reviewed 45 patients with peritonitis due to gastrointestinal perforation in patients with CAPD. The most common site of perforation was the sigmoid colon. Catheters were a rare cause of perforation, involving 3 cases. Takagi et al. [26] considered that the main cause of perforation was the addition of chronic catheter-related stimulation of the intestinal tract to underlying conditions of amyloidosis and diverticulitis. In our case, no catheter-related chronic inflammation, amyloidosis or diverticulitis was observed. The dialysis catheter and hematoma were clearly separate in this patient, with no adhesions between the catheter and surrounding tissue. The tip of the catheter was thus considered unlikely to have injured the sigmoid colon in this episode. In that sense, we ruled out involvement of the catheter.

Reddish drainage fluid from peritoneal dialysis is often related to menses or ovulation, particularly ovarian cyst rupture [27]. Although the reddish drainage fluid in the present case was due to intramural hematoma of the sigmoid colon fatally rupturing into the abdominal cavity, the patient had previously experienced reddish drainage fluid that had been attributed to menstrual issues.

Amyloidosis is a complication of dialysis and must therefore be considered. The prevalence of amyloidosis is similar in hemodialysis and peritoneal dialysis patients [28]. A case of gastric and colonic submucosal hematoma with amyloidosis has been reported, although that patient had not undergone dialysis [29]. Therefore, in the present case, we should consider the possibility that gastrointestinal amyloidosis occurred as a complication of dialysis and intramural hematoma occurred in the gastrointestinal tract. Although amyloidosis occurs in patients with end-stage renal failure on long-term CAPD [28], the patient in our case had a relatively short dialysis history of only 1 year, and amyloidosis due to dialysis would thus have been unlikely. In addition, amyloid deposition as seen in the above case report [29] was not observed microscopically in our case.

In conclusion, we have presented a rare case of ruptured idiopathic intramural hematoma of the sigmoid colon. Nonenhanced CT alone was able to identify the site of bleeding and matched well with findings from autopsy and pathology. With these findings on CT, intramural hematoma should be considered among the differential diagnoses.

Patient consent

Written consent was obtained from the patient's close relative for publication of the case information.

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