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Case Report

Spontaneous submucosal hematoma in the sigmoid colon causing complete Intestinal obstruction in a patient with cerebral palsy

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ABSTRACT

Background: Intestinal submucosal hematoma (ISH) is often associated with coagulopathy and anticoagulant drug use. ISH is frequently seen in the esophagus, duodenum, and rarely in small intestine in the gastrointestinal tract. It is very rare in the colon. In this case report, we present a 20-year-old male patient who developed intestinal obstruction due to spontaneous submucosal hematoma in the sigmoid colon. In the light of our English literature review, it was found that this was the first case in which spontaneous submucosal hematoma was detected in the sigmoid colon.

Case Report: In this case report, a 20-year-old male patient with a diagnosis of ileus with peroperative spontaneous submucosal hematoma was discussed. The patient had been suffering from inability to defecate and rectal bleeding with mucus for three days. The patient underwent laparotomy with the diagnosis of mechanical intestinal obstruction.

Conclusion: ISH is a clinical picture that frequently occurs due to anticoagulant use. It should be investigated in terms of ISH when prolonged INR is detected in elderly patients applying with abdominal pain. Surgical treatment should be performed in the presence of unimpaird intestinal obstruction or peritonitis.

KEYWORDS: hematoma, intestinal, obstruction, submucosal, spontaneous

INTRODUCTION

Intestinal submucosal hematoma (ISH) was first reported by McLauchlan in 1838^[1]. It is often associated with coagulopathy and use of anticoagulant drugs^[2,3]. Except for the use of anticoagulants, it may occur due to abdominal trauma, blood dyscrasias, and iatrogenic causes^[1,4-6]. In gastrointestinal tract, it is frequently observed in esophagus, duodenum, and rarely in the small intestine. It is very rare in the colon^[1,7]. The clinical symptoms typically include abdominal pain, intestinal obstruction and bleeding^[1,8]. ISH is a rare cause of intestinal obstruction^[2,9].

In this case report, a 20-year-old male patient who developed intestinal obstruction due to spontaneous submucosal hematoma in the sigmoid colon was examined. In the literature review, it was observed that this was the first case who had spontaneous submucosal hematoma in the sigmoid colon.

CASE REPORT

In this case report, we discussed a 20-year-old male patient who was diagnosed with ileus and who showed spontaneous submucosal hematoma during operation. From the anamnesis, it was understood that the patient received ventilatory support at home due to cerebral palsy. In this patient's history, no rectal enema was used the last 10 days. The patient had been suffering from intestinal gas-obstructed defecation and rectal bleeding with mucus for three days. Patient was under follow-up in the intensive care unit and in the physical examination of general surgery, diffuse abdominal distension was noted and discharge in the

form of strawberry jam was detected during rectal examination. Rectosigmoidoscopy was performed under sedation anesthesia in the intensive care unit. Rectosigmoidoscopy revealed necrotic mucosa and complete obstruction of the lumen at 15 cm from the rectum and there was no proximal transition. Figure 1 shows the endoscopic images of the patient.

Computed tomography (CT) of the abdomen was performed and it was observed that rectal contrast agent did not pass through the proximal part of sigmoid colon and that the intestinal loops were dilated from the proximal part of the sigmoid colon. Abdominal CT image of the patient is shown in Figure 2.

The patient underwent laparotomy due to diagnosis of mechanical bowel obstruction. During the operation, it was observed that all colon segments were severely dilated from the proximal part of the sigmoid colon and that intestinal segment, measuring 15cm, was necrotic and that its lumen was completely obstructed. The necrotic intestinal segment was totally excised up to the proximal and distal viability limits. During the operation, it was observed that necrosis and obstruction developed due to dissection associated with submucosal hematoma in the necrotic intestinal segment. After the resection, it was decided to perform end colostomy because of excessively dilated proximal colon segment. Distal segment was closed with stapler. Resected sigmoid colon segment are shown in Figure 3.

DISCUSSION

ISH is a rare condition that was first described by McLauchan in 1838; and non-traumatic ISH, on the other hand, was first described by Sutherland in 1904^[10]. It usually occurs in males and at an average age of 64. ISH occurs more frequently in the colon than the small intestine^[1]. Possible pathophysiology: The bleeding caused by the rupture of the terminal artery originating from the mesenteric artery causes the dissection between the muscularis mucosa and the muscular layer^[10]. The symptoms of ISH are nonspecific and may manifest with clinical symptoms ranging from mild abdominal pain to hemorrhagic shock^[11]. Often, the initial symptom is abdominal pain with nausea and vomiting. Gastrointestinal bleeding may occur as a result of rupture of the hematoma into the mucosa, or rupture into the abdomen may lead to peritoneal irritation^[10]. In this case report, the patient had the symptoms of rectal bleeding and abdominal distention.

ISH often occurs as a complication to high-dose anticoagulant therapy. Other risk factors include hemophilia, idiopathic thrombocytopenic purpura, leukemia, lymphoma, myeloma, chemotherapy, vasculitis, pancreatitis, pancreatic cancer, trauma, and iatrogenesis^[1,10]. In a study conducted by Kang *et al*^[12] in 2018, they reported that there was a history of anticoagulant/anti-platelet drug use in 78.6% of 103 small bowel-induced ISH cases in the literature in the last 30 years. In the same study, it was detected that over 90% of patients had underlying cardiovascular co-morbidity such as hypertension and cardiac arrhythmia^[12]. Yoldas *et al*^[10] reported that 85.7% of the patients had a history of anticoagulant/anti-platelet drug use, and two patients with no drug use had a history of hypertension. The patient in this case report

did not have anticoagulant / antiplatelet drug use and any trauma or comorbidity that could lead to bleeding. Although cases with colonic ISH due to an underlying disease, trauma or iatrogenic cause leading to anticoagulant use and coagulopathy have been reported in the literature, in the light of literature reviews, it was found that this patient was the first patient to have spontaneous ISH in the sigmoid colon without any use of anticoagulant/anti-platelet drug or any underlying cause^[8,13-15].

If a patient presents with the complaint of abdominal pain and he/she has history of prolonged International Normalized Ratio (INR), ISH should be considered^[10]. As the patient in this case report had no history of anticoagulant use or an underlying disease that could lead to coagulopathy, no prolongation in INR was detected. In the diagnosis of ISH, barium enemas, ultrasonography (USG) and CT are used. USG sensitivity is reported as 71.4% and CT sensitivity as 80 - 100%. Among these, CT is the most useful examination. In addition, if the patient is stable and the diagnosis is not clear, endoscopic examination for tumor and hemorrhage discrimination can be performed^[1,10]. In this case report, rectosigmoidoscopy was performed because of rectal bleeding, and mucosal necrosis and complete obstruction was observed in the lumen.

There are not enough studies for standard therapy of ISH. However, early diagnosis is very important for the success of medical treatment response^[10]. The conservative approach is the first choice of treatment. In conservative treatment, anticoagulant / antiplatelet medication should be discontinued first. Coagulation parameters should be corrected with vitamin K and fresh frozen plasma, oral intake should be discontinued and nasogastric decompression should be performed^[1,10]. In hemodynamically stable patients, if there is obvious arterial bleeding, endovascular methods should be preferred^[15]. Surgical treatment is recommended in patients with hemodynamic instability, ineffective intestinal obstruction and intestinal necrosis or signs of peritonitis^[8,15]. In this case report, the patient underwent surgical treatment because of the detection of mucosal necrosis and obstruction endoscopically, and the rectal contrast agent could not pass to proximal intestinal loop in CT. Resection+anastomosis or resection+end colostomy can be performed in surgical treatment^[16]. In this case report, end-colostomy was performed as the intestinal loops in the proximal part were excessively dilated after segmental colon resection. Recently, several new treatment strategies have been reported as alternative to surgical treatment. These are percutaneous drainage under USG guidance and endoscopic incision and drainage. However, it should be known that there is a risk of intestinal perforation in these two treatment modalities. As the patient in this case report had total intestinal obstruction that prevented endoscopic passage, incision and drainage could not be performed^[1,10].

CONCLUSION

ISH is a clinical picture that frequently occurs due to use of anticoagulants. For elderly patients who present with the complaint of abdominal pain, ISH should be considered, if

prolonged INR is detected. However, as in this case report, it should be kept in mind that ISH and intestinal obstruction may develop without any underlying cause. Conservative treatment is the preferred choice and early diagnosis increases the success of conservative treatment. Surgical treatment should be performed in the presence of unimpaired intestinal obstruction or peritonitis. Current treatment modalities, such as percutaneous drainage and endoscopic drainage with USG, should be considered as an alternative to surgery, but the risk of perforation should be taken into consideration.

ACKNOWLEDGMENTS

Author Contributions

Investigation: Ugur KESICI, Sevgi KESICI

Methodology: Ugur KESICI

Validation: Ugur KESICI, Sevgi KESICI

Writing – original draft: Ugur KESICI

Writing – review & editing: Ugur KESICI, Sevgi KESICI

I and all author have no conflict of interest and have no financial support

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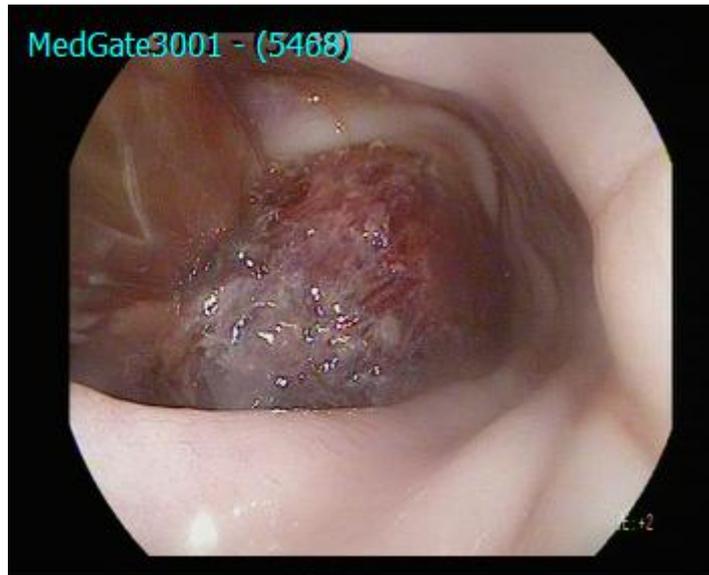


Figure 1: Endoscopic image of the patient

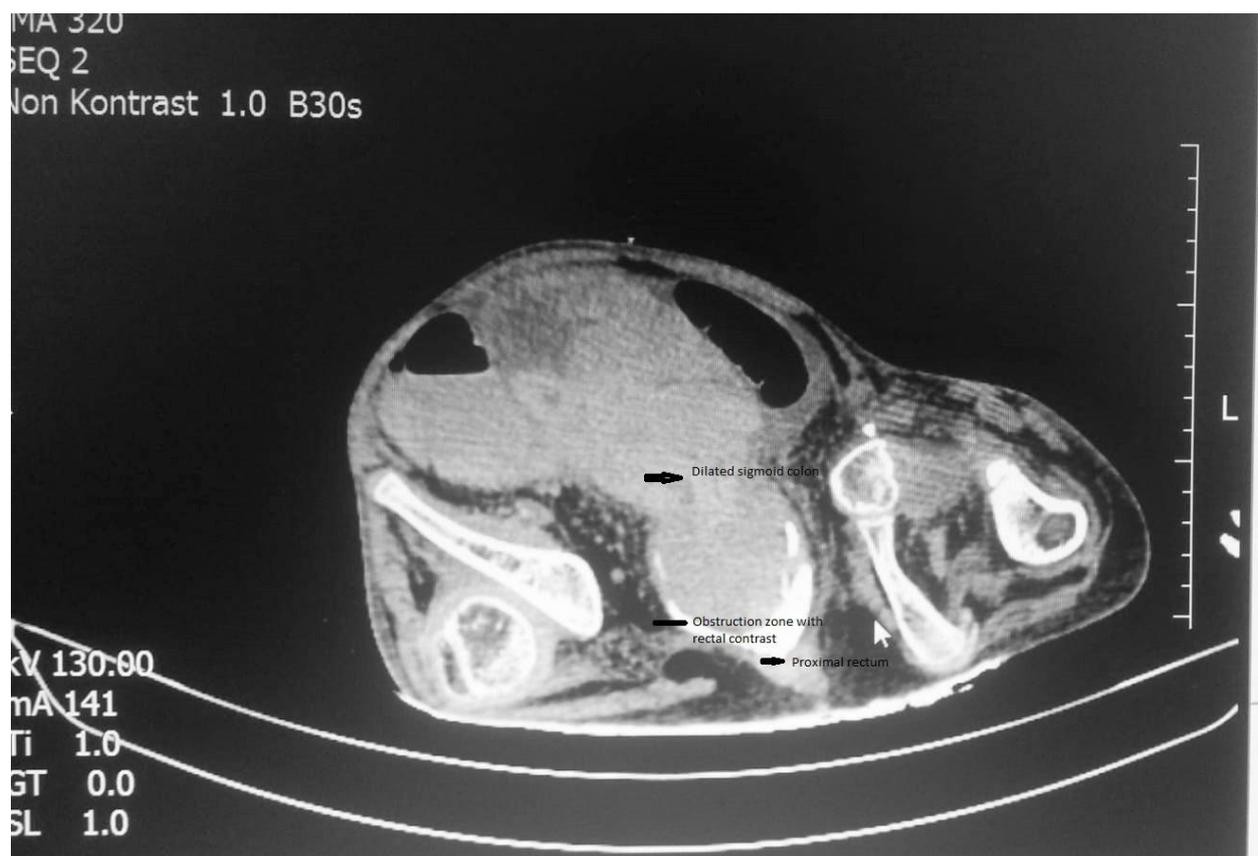


Figure 2: Abdominal CT image of the patient



Figure 3: Image of resected colon segment