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Klinik Çalışma

Spontaneous intramural hematoma of the small intestine

Spontan intramural ince bağırsak hematomu

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BACKGROUND
Spontaneous intramural hematoma of the small intestine is a rare clinical condition that may result in potentially serious complications. The purpose of this study was to present our experience with the diagnosis and management of spontaneous intramural hematoma of the small intestine.

METHODS
The medical records of the patients with spontaneous intramural hematoma of the small intestine were retrospectively reviewed. Six patients were included in this study.

RESULTS
Anticoagulation therapy and factor VIII deficiency were found to be responsible for the intramural hemorrhage in five patients (83%) and one patient, respectively. Acute abdominal pain followed by nausea and vomiting were the most common presenting symptoms. Abdominal computed tomography scan was diagnostic in five of the six patients. Four patients were followed up with conservative therapy. Surgical intervention was required in two patients due to acute abdomen. All patients were discharged from the hospital uneventfully.

CONCLUSION
The patient's medical history, physical examination and radiological evaluation proved adequate for the diagnosis. Conservative therapy provides regression of the hematoma in most patients. Surgery should be reserved only for the complicated cases.

Key Words: Anticoagulant; hematoma; hemophilia; small intestine; intramural hematoma.

AMAÇ
Spontan ince bağırsak hematomu nadir görülen ve ciddi komplikasyonlara neden olabilen bir klinik durumdur. Bu çalışmanın amacı, spontan intramural ince bağırsak hematomu saptanan olgularda tanı, tedavi ve takipteki tecrübelerimizi sunmaktır.

GEREÇ VE YÖNTEM
İnce bağırsak spontan intramural hematom tanısı konulan hastaların verileri retrospektif olarak incelendi. Altı hasta çalışmaya dahil edildi.

BULGULAR
İntramural kanamaya neden olarak dört hastada (%83) antikoagülasyon tedavisi ve bir hastada faktör VIII eksikliği belirlendi. Akut karın ağrısı, bulantı ve kusma en sık başvuru şikayetleri idi. Altı hastanın beşinde tanı karın bilgisayarlı tomografisi ile kondu. Dört hasta konservatif olarak takip edildi ve iki hastada akut karına bağlı cerrahi girişime gerek duyuldu. Tüm hastalar sorunsuz taburcu edildi.

SONUÇ
Fiziksel inceleme ve radyolojik tetkikler hastaların özgeçmişleri ile beraber değerlendirildiğinde tanı için yeterlidir. Konservatif tedavi hastaların çoğunda hematomun gerilemesini sağlar. Cerrahi tedavi komplike olgularda uygulanmalıdır.

Anahtar Sözcükler: Antikoagülan; hematom; hemofili; ince bağırsak; intramural hematom.

Intramural small intestinal hematoma, once considered a rare clinical entity, is being reported with increasing frequency. It was first reported by McLouchlan in 1838, and the first radiological description regarding jejunal hematoma was given by Liverud more than 100 years later.[1] Abdominal trauma is the most common etiological factor. Non-traumatic cases are referred to as spontaneous intramural hematoma of the small intestine (SIHSI), in which overanticoagulation is the most common cause. Other conditions associated with SIHSI are bleeding disorders (e.g. hemophilia and idiopathic thrombocytopenic purpura), malignancies and vasculitis.[2-4] The presentation of patients with SIHSI can vary from mild and vague ab-

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dominal pain to intestinal tract obstruction and acute abdomen.[1,3-8] Intraperitoneal hemorrhagic effusion can also be present. The diagnosis is often not suspected and usually established after radiological imaging or surgical exploration.[4]

In this report, we aimed to present our experience in patients with SIHSI, and to document the etiological factors, clinical presentations, diagnosis, and management of this rare condition.

MATERIALS AND METHODS

This study includes the patients who were consecutively registered at Istanbul University, Cerrahpasa Medical Faculty, Emergency Surgery Unit between October 2003 and October 2009 and diagnosed as having SIHSI. The medical records of these patients were analyzed retrospectively. Patient demographic data (sex, age at diagnosis of SIHSI), the coexisting medical conditions, previous medications, presenting symptoms and signs, laboratory and radiological findings, management interventions, hospitalization course, and patient outcome were evaluated.

RESULTS

Six patients were included in this study. All the patients were male, with an average age of 55 years (range: 24-76 years). Patient demographic data and the clinical characteristics are summarized in Table 1.

Patient past medical history questionnaire revealed anticoagulation medication with warfarin alone or together with acetylsalicylic acid due to coexisting medical conditions in five patients, cardiovascular diseases in four patients (67%), and cerebrovascular accident in one patient. One patient had factor VIII deficiency (Case 5).

Acute blunt abdominal pain followed by nausea and vomiting were the most common presenting symptoms. The mean duration of the symptoms was 2.5 days (range: 1-5 days) prior to hospital admission. On physical examination, two of six patients had guarding and rebound tenderness, consistent with acute abdomen (Cases 2 and 3). There was no hemodynamic instability in any case.

On admission, the laboratory results indicated leukocytosis in five patients (83%), with a mean leukocyte count of 18,700/mm^3 (range: 14,600-22,700/mm^3). Four patients were anemic (hemoglobin level ≤13.6 g/dl). Increased international normalized ratio (INR) values were detected in all patients, being unmeasurably high in three patients.

Radiological studies included plain abdominal radiography, ultrasonography (US) and computed tomography (CT) scan. Small bowel obstruction was observed

Table 1. The demographic data and the clinical features of the patients diagnosed as having spontaneous intramural hematoma of the small intestine

No	Age /Sex	Past medical history	Previous medication	Physical signs	Laboratory findings	Radiological findings	Treatment	Hospital stay (d)
1	24/M	AVR due to aortic valve disease	Warfarin	Epigastric tenderness	WBC: 15,000/mm^3 Hb: 15 mg/dl aPTT: 142.7 sec PT: >60 sec INR: *	X-ray: air-fluid levels US: inconclusive CT: segmental thickening of the jejunal wall resulting in partial bowel obstruction	Conservative	4
2	55/M	Stroke and coronary by-pass due to atherosclerosis	Warfarin	Guarding and rebound tenderness	WBC: 21,000/mm^3 Hb: 14.3 mg/dl aPTT: 112 sec PT: 80.4 sec INR: 8.5	X-ray: normal CT: diffuse jejunal wall thickening on a 25 cm segment suggesting mesenteric ischemia and massive pelvic fluid	Surgery (1: diagnostic laparoscopy + drainage, 2: second-look laparoscopy)	12
3	72/M	Intracardiac thrombosis + peripheral atherosclerosis	Warfarin + acetyl-salicylic acid	Guarding and rebound tenderness	WBC: 22,700/mm^3 Hb: 12 mg/dl aPTT: >120 sec PT: >120 sec INR: *	X-ray: air-fluid levels CT: multisegmental diffuse jejunal and ileal wall thickening each measuring up to 10 cm in length and hemoperitoneum	Surgery (1: diagnostic laparoscopy → conversion + drainage, 2: second- look laparotomy)	14
4	69/M	Coronary by-pass due to atherosclerosis	Warfarin + acetyl-salicylic acid	Generalized abdominal tenderness	WBC: 14,600/mm^3 Hb: 10.5 mg/dl aPTT: 154 sec PT: 116 sec INR: 7.93	X-ray: air-fluid levels US: jejunal wall thickening CT: segmental jejunal wall thickening and moderate perihepatic effusion	Conservative	9
5	76/M	Cerebrovascular ischemia due to chronic atrial fibrillation	Warfarin	Abdominal distension and tenderness, melena, macroscopic hematuria	WBC: 20,200/mm^3 Hb: 12.7 mg/dl aPTT: >120 sec PT: >60 sec INR: *	X-ray: air-fluid levels CT: mural wall thickening of the distal ileum and bilateral renal pelvic hematoma	Conservative	13
6	37/M	Factor VIII deficiency	–	Generalized abdominal tenderness	WBC: 10,700/mm^3 Hb: 9.7 mg/dl aPTT: * PT: 12.6 sec INR: 1.07	X-ray: normal US: normal CT: distal ileal wall thickening	Conservative	10

AVR: Aortic valve replacement; WBC: White blood cell; Hb: Hemoglobin; aPTT: Activated partial thromboplastin time; PT: Prothrombin time; INR: International normalized ratio; *: unmeasurably high; d: Days; M: Male.

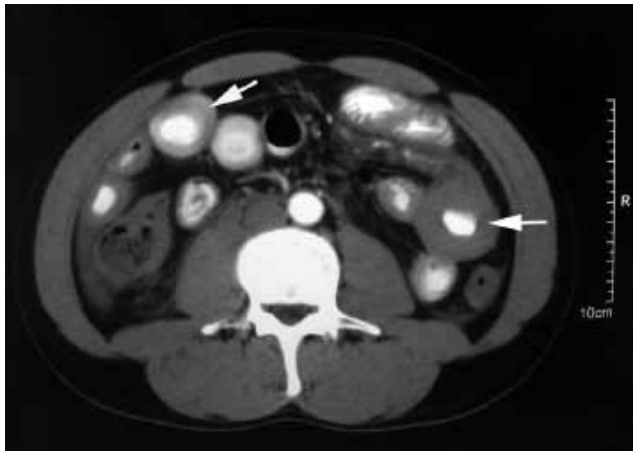

Fig. 1. CT scan of the upper abdomen with oral contrast demonstrates homogeneous and concentric wall thickening in a long jejunal segment (arrows) (Case 1).

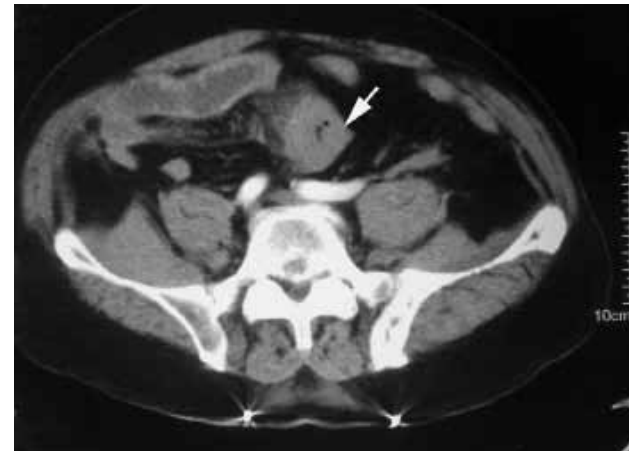

Fig. 3. CT scan of the lower abdomen demonstrates concentric mural wall thickening of the distal ileal segment (arrow) (Case 5).

in three patients (50%). US was diagnostic in one patient. CT scan with or without oral contrast administration was found to be a preferred modality, being diagnostic in five cases (83%) (Figs. 1-4). CT demonstrated intramural wall thickening involving the jejunum in four patients, the ileum in one patient, and both in one patient (Case 3). A single hematoma was noted in five patients and multiple hematomas in one. The involved small bowel segments, estimated from the CT scan, had a mean length of 17 cm (range: 10-20 cm).

Four patients (66%) were managed conservatively. Conservative therapy included bowel rest, cessation of anticoagulant and antiplatelet drug administration, fresh frozen plasma (FFP) (range: 2-8 units), packed red blood cells (range: 1-5 units), factor VIII concentrates, and vitamin K as required. Surgical intervention, in addition to medical therapy, was performed in the remaining two patients in whom the clinical and CT findings were suggestive of acute abdomen: for a possibility of mesenteric ischemia (Case 2) and the presence of a massive intraperitoneal hematoma (Case 3). However, none required a bowel resection. Nasogastric tube decompression was applied in all patients.

All patients responded well to the treatment and were discharged uneventfully. The mean length of hospital stay was 10.3 days (range: 4-14 days). At discharge, patients who had been on anticoagulation treatment previously resumed warfarin at therapeutic doses. Follow-up CT scans obtained from two patients demonstrated regression of the hematoma. There were no further short-term complications on follow-up visits.

DISCUSSION

Despite its rarity, SIHSI has become increasingly recognized both because of the availability of current radiological diagnostic methods and the increasing number of published papers regarding this condition as a complication of anticoagulation therapy and bleeding disorders.[1,3,4,6-10]

The most probable physiopathology of SIHSI would be characterized by the shredding of the terminal arteries as they leave the mesentery and penetrate the muscularis layer of the intestinal wall. Consequently, the hemorrhage dissects the bowel wall between the muscularis mucosa and the muscularis layers. Unlike mesenteric vascular occlusion, the vi-

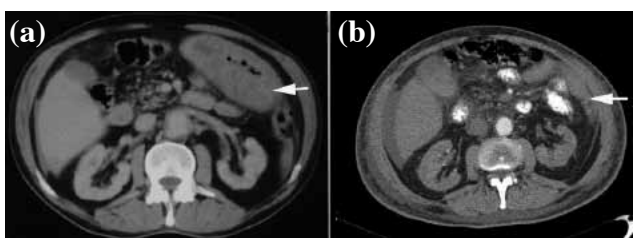

Fig. 2. Intravenous contrast-enhanced CT scan demonstrates diffuse but eccentric intramural jejunal wall thickening for a length of 20 cm as a mass compressing the lumen (arrow) and there is moderate perihepatic effusion **(a)**. Follow-up CT scan with oral contrast after 10 days shows the regression of the hematoma **(b)** (Case 4).

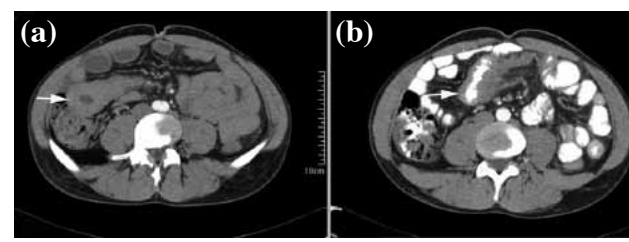

Fig. 4. CT scan of the lower abdomen without oral contrast demonstrates diffuse and concentric ileal wall thickening (arrow) for a segment of 11 cm in a patient with factor VIII deficiency on admission **(a)**. After 10 days, follow-up CT scan with oral contrast demonstrates a decrease in the mural wall thickness of the involved ileal segment **(b)** (Case 6).

ability of the mucosa is preserved.[5,11,12] As shown in our results, the clinical condition may vary, and the symptoms and signs of the small bowel obstruction or acute abdomen may predominate. It was theorized that the progression of the symptoms was due to insidious bleeding in the submucosal layer and simultaneous formation of an intramural osmotic gradient leading to an expansion.[3] Intraperitoneal hemorrhagic effusion, when present, is related to the leakage of blood from an engorged, thickened and inflamed bowel wall with submucosal bleeding into all layers.[3,13,14]

Once SIHSI is suspected, the patient's past medical history and current medication should be carefully questioned in order to establish the correct diagnosis and management. Oral anticoagulation therapy by warfarin was reported to be the most common predisposing factor for SIHSI.[1,3,4] The incidence of this condition among patients receiving this therapy is 1 in 2500.[2] According to a review report presented by Sorbello et al.,[1] the incidence was higher in males, and the age of patients varied from 32 to 78 years. Hemophilia A, which is characterized by a lack of coagulation factor VIII, is another predisposing factor for SIHSI.[13] Katsumi et al.[15] reviewed 30 hemophilia A patients since 1964 who suffered from intramural hematoma of the gastrointestinal tract. Their ages ranged from 8 to 78 years, and small intestine involvement was found in 13 patients. However, the incidence of SIHSI in hemophiliacs, in general, remains unknown. In our study, with a relatively limited number of patients, SIHSI was associated with previous anticoagulant medication in the majority of the patients.

Routine laboratory tests in cases suspected for SIHSI should include complete blood count, routine biochemical analysis, coagulation parameters, and bleeding time. INR is used as an indicator of the anticoagulant effect, and values greater than 3 are associated with an increased risk of bleeding. Bleeding can also occur even if the coagulation parameters are within the therapeutic range.[6,8] In the present study, the majority of patients had leukocytosis and were anemic on admission. The INR levels of three patients were detected to be unmeasurably high. With appropriate medication, these parameters in all the cases were noted to be in the normal range prior to hospital discharge.

Plain abdominal radiography is seldom specific and may not show evidence of intestinal obstruction. The barium X-ray studies were widely used until the 1980s; however, they lost ground as a diagnostic method with the development of US and CT. These methods offer a more adequate evaluation regarding the presence or absence of free fluid in the abdominal cavity. Despite the diagnostic capability of US, CT is the diagnostic modality of choice and it has proven to be a sensitive method, defining the site and the extent of hemorrhage as "coiling spring" and "pseudo-kidney" signs.[1,8] Through the gastrointestinal tract, the jejunum is the most affected region in up to 70% of such cases.[1,3] Our radiological findings, combined with the clinical presentation, were sufficient to make a confident diagnosis in the majority of the patients. The average length of the involved bowel segment was 17 cm, which had been reported to be 23 cm in a study concerning 13 patients.[3] Partial resolution of the hematoma was noted on follow-up CT on the 10th day after onset at the earliest. A few authors have suggested that non-contrast CT scan should be performed prior to the routine oral and intravenous contrast-enhanced scan, because they claim that contrast-enhanced scan alone may mask the presence of intramural hemorrhage.[1,3,16] However, in our series, the use of contrast medium had no crucial role in revealing SIHSI, and all the CT scans were diagnostic regardless of the methods used.

Non-operative medical treatment is usually the initial approach for SIHSI.[1,8,13,15] Surgical exploration, which was previously used as a diagnostic method, should be reserved for patients in the evidence of complications such as suspected ischemia with or without bowel perforation, peritonitis, intraabdominal hemorrhage, and intractable intestinal obstruction.[1,13] In the case of anticoagulant-induced intramural intestinal hematoma, bowel rest, nasogastric decompression, correction of electrolyte disturbances, blood transfusion, and correction of coagulopathy with FFP and vitamin K are the mainstays of therapy. It appears to be safe to continue anticoagulation therapy within the therapeutic range after the resolution of the hematoma.[7] If the underlying pathology is factor VIII deficiency, most of these intramural hematomas can be managed with simple non-surgical and conservative intervention, as long as the bleeding can be controlled by prompt and vigorous factor VIII replacement.[13]

The clinical and CT findings of acute abdomen prompted us to perform a surgical intervention in two patients. We performed diagnostic laparoscopy in one patient (Case 2), as the preoperative findings suggested mesenteric emboli. On laparoscopy, the bowel segments were found to be normal except for a wall thickening in the jejunal region 15 cm distal to the ligament of Treitz and oozing type of bleeding from the serosa. At the aforementioned region, the bowel took on a dark purple color due to the diffuse mural hematoma. Free pelvic hemorrhagic fluid was suctioned and a 10-mm trocar was kept in place at the right lower quadrant in order to perform a second-look laparoscopy. The next day, the hematoma was observed to be nonexpanding; therefore, the procedure was terminated and the trocar was removed. In the other patient (Case 3), we converted the laparoscopic procedure to open surgery due to the presence of massive intraperi-

toneal hemorrhage. Subsequent to the suction of 1000 ml of blood, we noted the presence of multisegmental small bowel hematomas and intraluminal blood in the transverse colon with no necrosis of the bowel wall. Second-look laparotomy was performed the next day, which revealed gradual regression of the hematoma.

In uncomplicated cases where surgery is not required, regression of SIHSI tends to occur within a week,[1,3] and complete resolution usually occurs within two months after onset. Persistent lesions lasting more than two months should be further investigated for other pathologies.[1,17]

In conclusion, SIHSI is a potentially serious condition with different underlying etiologies and various clinical presentations. A high index of suspicion should be maintained in patients with acute abdominal pain who receive anticoagulation therapy or have any bleeding disorders. Early diagnosis and prompt medical treatment are important in order to avoid unnecessary surgical interventions. Surgery should be reserved for complicated cases or when the diagnosis is uncertain.

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