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A case of antiphospholipid antibody syndrome with Budd-Chiari and colonic ulcers complicated with gastrointestinal hemorrhage

Budd-Chiari ve kolonik ülserlerle seyreden antifosfolipid antikor sendromu vaka sunumu

Özer İLKGÜL[1], Gökhan İÇÖZ[2], Murat DAYANGAÇ[2], Yaman TOKAT[2], Ömer ÖZÜTEMİZ[3]

Celal Bayar University Medical Faculty, Department of Surgery[1], Manisa
Ege University Medical Faculty, Department of Surgery[2], Department of Gastroenterology[3], Izmir

The antiphospholipid antibody syndrome (APA) is characterized by an increased incidence of venous and arterial thrombosis. APA syndrome has some gastroenterological manifestations such as Budd-Chiari syndrome, hepatic infarction, esophageal necrosis, intestinal ischemia, pancreatitis and colonic ulceration. We report a 34-year-old man with APA syndrome complicated by hepatic venous thrombosis (Budd-Chiari) and colonic ulcers. The clinical and laboratory findings were compatible with APA syndrome that developed secondary to systemic lupus erythematosus. In order to initiate anticoagulant therapy, he was heparinized. Since lower gastrointestinal bleeding developed, heparin was discontinued and the patient was followed up with baby aspirin and steroids. This case report extends the gastroenterological manifestations of the APA syndrome to include colonic ulceration, which may outweigh the efficacy of initial anticoagulant therapy.

Key words: Antiphospholipid antibody syndrome, systemic lupus erythematosus, colonic ulcers

Antifosfolipid antikor sendromu (AFA), artmış venöz ve arteriyel tromboz insidansı ile karakterize bir sendromdur. AFA sendromununi; Budd-Chiari sendromu, hepatik iskemi, özofagus nekrozu, intestinal iskemi, pankreatit ve kolon ülserleri gibi gastrointestinal sistem yansımaları bulunmaktadır. Burada; hepatik venöz tromboz (Budd-Chiari) ve kolon ülserleri ile karakterize AFA sendromlu 34 yaşında erkek olgu sunuldu. Olgunun klinik ve laboratuar bulguları SLE'ye sekonder gelişen AFA sendromu ile uyumlu idi. Hastaya antikoagülan tedavi amacıyle heparinizasyon uygulandı. Alt gastrointestinal sistem kanaması gelişmesi üzerine heparinizasyon sonlandırıldı ve yerine aspirin ve steroid tedavisi başlandı. Bu olgu; AFA sendromunun kolonik ülserlerle seyreden ve antikoagülan tedavi etkinliğinin fazla oranda gözlendiği bir olgudur.

Anahtar kelimeler: Antifosfolipid antikor sendromu, sistemik lupus eritematozis, kolonik ülserler

CASE REPORT

A 34-year-old man was admitted to hospital with abdominal pain, jaundice and weight loss. He had a history of laparotomy for acute abdomen a month before the admission. The liver biopsy revealed only ascites and hepatomegaly. Histopathological examination showed centrilobular necrosis. For further evaluation, duplex ultrasonography and angiography were performed, which showed right hepatic venous thrombosis only. He then underwent angiographic balloon dilatation (Figure 1). Following angiography, he was heparinized. Two days later, he had a lower gastrointestinal bleeding, for which heparin was discontinued. Colonoscopic examination revealed multiple colonic ulcers in descending colon, and biopsies were taken (Figure 2). He was followed up with baby aspirin and steroids. Colonoscopic biopsy samples showed ischemic ulcers. Meanwhile, laboratory tests showed elevated liver enzymes with thrombocytopenia and prolonged partial thromboplastin time; however, antithrombin III, and proteins C and S were within the normal range. Anti-nuclear antibody (ANA) (1/1280 peripheric pattern) and anti-cardiolipin antibody (ACA) (ACA Ig G>120)

Address for correspondence: Özer İLKGÜL
Didim: 2. Giriş: 7-8, D: 4, Atakent-Karşıyaka, Izmir, Turkey
Phone: +90 232 330 99 81
Gsm no: +90 532 775 04 40
E-mail: ilkgul@doctor.com

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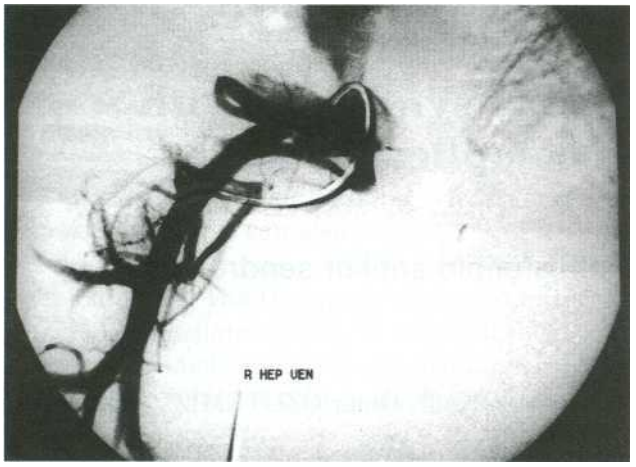

Figure 1. Angiographic balloon dilatation of hepatic vein

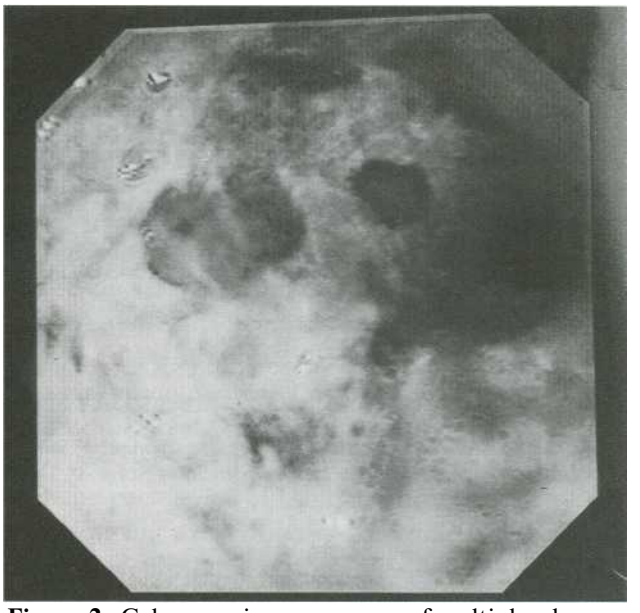

Figure 2. Colonoscopic appearance of multiple ulcers

were positive. The clinical and laboratory findings were compatible with antiphospholipid antibody (APA) syndrome in this patient. APA syndrome may often be associated with connective tissue diseases, especially systemic lupus erythematosus (SLE), but in this case laboratory findings (peripheric pattern of ANA) just supported the diagnosis of SLE, rather than indicating it distinctly.

DISCUSSION

Antiphospholipid antibody syndrome is characterized with frequent thromboses, repeated fetal losses and thrombocytopenia, and may develop in association with SLE (1). The clinical manifestations of APA syndrome include migraine, chorea, hemolytic anemia, heart valve disease, malignant hypertension and pre-eclampsia, as well as the gastroenterological manifestations, i.e. hepatic infarction, esophageal necrosis, intestinal ischemia, pancreatitis, Budd-Chiari syndrome and colonic ulcers (1, 2). APA syndrome may develop primarily or in association with a variety of clinical conditions, such as autoimmune disorders, infectious diseases, and neoplastic disorders (secondary APA syndrome-SAPS) (2, 3). Long-term anticoagulant treatment is preferred to decrease the complication rates (4, 5, 6). However, anticoagulant treatment may deteriorate the clinical outcome in patients with gastrointestinal ulcers, as was seen in our case.

This case report extends the gastroenterological manifestations of the APA syndrome to include colonic ulceration, which may outweigh the efficacy of initial anticoagulant therapy.

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