

## Giant peritoneal cyst hydatid and albendazole-induced aplastic anemia in noncirrhotic portal hypertensive patient

Nonsirotik portal hipertansiyonlu hastada dev peritoneal kist hidatik ve albendazole bağı aplastik anemi

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*Albendazole has broad-spectrum coverage as an antiparasitic drug, and the reported side effects have been minimal. We report the case of a patient with febrile neutropenia beginning during the second week of therapy for a hepatic, splenic and peritoneal echinococcal cyst. This case was a 49-year-old man who presented with a large cystic abdominal mass. His medical history was significant for non-cirrhotic portal hypertension and recurrent cholangitis. Albendazole sulfoxide peak dose and half-life are significantly prolonged by liver disease and concomitant administration of certain drugs. The severity and duration of albendazole-induced neutropenia in this case was likely related to the underlying liver disease. Frequent serial monitoring of blood counts and cessation of medication with any evidence of marrow toxicity in such patients are warranted.*

**Key words:** Cyst hydatid, albendazole

### INTRODUCTION

Hydatidosis is an endemic illness in regions of the Middle Orient, Mediterranean, south of America, North Africa, and Australia. The preferential localization of cyst hydatid is the liver (48%) and lung (36%), but in 6% of cases, rare localizations, such as the brain, are seen (1). The hepatic hydatid cyst is a major health problem in endemic areas. Hydatid disease of the liver is still endemic in certain parts of the world. The modern treatment of hydatid cyst of the liver varies from surgical intervention to percutaneous drainage or medical therapy. Surgery is still the treatment of choice and can be performed by the conventional or laparoscopic approach (2,3).

Albendazole is the drug of choice in the medical treatment of hydatidosis. It has a broad-spectrum coverage as an antiparasitic drug, and the reported side effects have been minimal (4). We report the case of a patient with febrile neutropenia beginning during the second week of therapy for a hepatic, splenic and peritoneal ec-

*Albendazol geniş spektrumlu antiparazitik bir ilaçtır. Yan etkilerinin minimal olduğu bildirilmiştir. Hepatik, splenik ve peritoneal ekinokokkal kist nedeniyle albendazol başlanan, ikinci haftada febril nötropeni gelişen bir vaka sunulmuştur. 49 yaşında erkek hasta karında büyük kistik kitleleri nedeniyle müracaat etti. Hastanın hikayesinde nonsirotik portal hipertansiyon ve tekrarlayan kolanjit atakları dikkat çekmekte idi. Albendazol sulfoksit peak dozu ve yarılanma ömrü karaciğer hastalıklarında ve başka ilaçlar kullanıldığında artmaktadır. Bu vakada ciddi nötropeni gelişmiş olmasının altta yatan karaciğer hastalığına bağlı olduğu düşünülmüştür. Albendazol başlanacak bu tür hastalar kemik iliği toksisitesi yönünden sık kan sayımları ile yakından takip edilmeli, verilecek diğer ilaçlara dikkat edilmelidir.*

**Anahtar kelimeler:** Kist hidatik, albendazol

hinococcal cyst. This case was a 49-year-old man who presented with a large cystic hepatic and splenic mass.

### CASE REPORT

A 49-year-old male patient followed in our outpatient clinic with non-cirrhotic portal hypertension (due to paroxysmal nocturnal hemoglobinuria), hydatid cyst and recurrent cholangitis was admitted with fever, chills, abdominal pain, and abdominal distension. The size of the hydatid cyst had increased from 10 cm to 20 cm (on his magnetic resonance imaging [MRI] studies 1 year later). The cyst was drained and 150 cc pure alcohol was injected. Albendazole 800 mg/day p.o. treatment was started, and the patient was discharged. On the 9<sup>th</sup> day of the medical treatment, he had malaise, fever and chills and was admitted to the emergency department. His white blood cell (WBC) count was 300/mm<sup>3</sup> and polymorphonuclear leukocytes (PNL) were 0/mm<sup>3</sup>.

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Febrile neutropenia was diagnosed and ampicillin-sulbactam treatment was started. He was later admitted to the Gastroenterohepatology inpatient service. His physical examination was not remarkable except for massive splenomegaly and palpable mass in the midline. His laboratory results showed agranulocytosis, total bilirubin 4.55 mg/dl, direct bilirubin 2.91 mg/dl, albumin 2.38 g/L, and gamma globulin 1.99 g/L. His blood smear was remarkable for the absence of neutrophils. Bone marrow aspiration showed loss of myeloid cells and was diagnosed as agranulocytosis due to albendazole. Albendazole treatment was stopped immediately and granulocyte colony-stimulating factor (G-CSF) (Neupogen™) 48 MU/day treatment was started. On the 5th day of the treatment, his WBC count was 5300/mm<sup>3</sup> and PNL were 2300/mm<sup>3</sup>.

His fever continued and the antibiotic was switched to piperacillin-tazobactam 4.5 g, p8d, i.v. according to blood culture results. In his abdominal MRI, perihepatic thick-walled new fluid collection was observed. Approximately 200 cc purulent material was drained and sent for culture. The hydatid cyst was found to be full again and was drained. Approximately 1500 cc bile-containing material was drained and sent for culture (Figures 1 A-B). Culture results showed *Escherichia coli* and *Klebsiella*, and the antibiotic was switched to imipenem-cilastatin 500 mg, q6d, i.v. After the 2nd day of the drainage, his fever resolved.

After the 14<sup>th</sup> day of the treatment, parenteral treatment was switched to moxifloxacin 400 mg, pd, p.o. He was discharged with antibiotic, ursodeoxycholic acid and propranolol treatments.

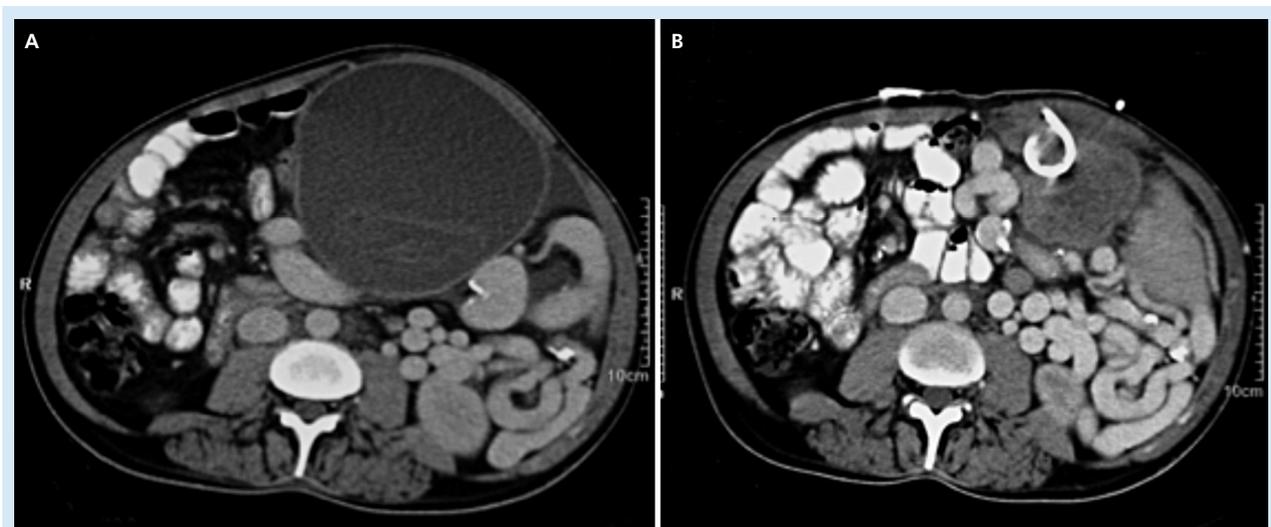
## DISCUSSION

The preferential localizations of cyst hydatid are the liver and lung (1). Other rare localizations are renal, cardiac, pancreas, ovarian, musculoskeletal, splenic, and intracranial (5-11). Peritoneal hydatidosis is another rare localization of hydatid disease, most often secondary to a hydatid cyst of the liver (12). In our patient cyst, localizations were hepatic, splenic and peritoneal.

In the assessment of liver hydatid cyst cases seen over 10 years in Turkey, in total, 69% of cases had a single cyst and 31% multiple cysts. The most common symptom was abdominal pain in 74% of patients. Right lobe involvement was encountered in 65% of cases, left lobe in 13%, and left and right in 8%. In 27% of the patients, cholelithiasis was the most common accompanying disease (13). In another study, single cysts were found in 65.7% of the cases, two cysts in 20% and multiple cysts in 12.5% (14). Our patient had three cysts, in three different areas.

Histology revealed parasites in the pericyst in 129 cases (34%), and a fistula from the cyst to the bile duct was observed in 47 cases (12.5%) (14). Our case had fistula from the peritoneal cyst to the bile duct and cutaneously.

The modern treatment of hydatid cyst of the liver varies from surgical intervention to percutaneous drainage or medical therapy. Surgery is still the treatment of choice (2). Our case with hydatid cyst in the liver associated with portal hypertension was not suitable for surgical treatment. Percutaneous drainage and alcohol injection were performed.



Figures 1. Giant peritoneal cyst hydatid (A) before and (B) after drainage.

Percutaneous drainage with alcohol injection for hydatid cysts has been commonly used in the last two decades. Albendazole is the drug of choice in the medical treatment of hydatidosis (4). Percutaneous drainage with alcohol injection was used in our patient first and was followed with albendazole treatment.

Albendazole has broad-spectrum coverage as an antiparasitic drug, and the reported side effects have been minimal. We report the case of a patient with febrile neut-

ropenia beginning during the second week of therapy for hepatic, splenic and peritoneal echinococcal cysts. This case was a 49-year-old man who presented with a large cystic peritoneal, hepatic and splenic mass.

In conclusion, the half-life of albendazole sulfoxide is increased in liver diseases. In this case, portal hypertension may have had an additive role in the occurrence of neutropenia. Such patients who will be on albendazole treatment should be followed with complete blood count tests frequently.

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