Anaphylaxis Due To A Spontaneously Ruptured Hydatid Cyst: A Case Report
S Devrim, M Gura, E Koltka, S Yuzer

Citation

Abstract
Spontaneous rupture of liver hydatid cyst causes an acute abdomen and leakage of a cyst contents into the blood circulation is a triggering factor for anaphylaxis. Abdominal ultrasonography and computed tomography are the first-line imaging studies in cases of suspected rupture of hydatid cyst. Urgent treatment should be initiated, relying first on emergency management of anaphylactic shock and later on surgical treatment of the cysts. We report a case of an anaphylaxis due to a spontaneously ruptured hydatid cyst.

INTRODUCTION
Hydatid disease is a parasitic infection caused by Echinococcus granulosus characterised by cyst formation in any organ, although the liver is the most commonly involved. Up to one third of patients with hepatic hydatid disease present with complications such as rupture (into the biliary tree, thorax or peritoneum), secondary infection, anaphylactic shock, sepsis and liver replacement. Anaphylactic reactions, which sometimes are the first manifestations of the disease, including urticaria, edema, respiratory symptoms, and anaphylactic shock due to spontaneous or provoked rupture of the parasite cyst. However, neurological symptoms such as fokal deficits and seizures are observed in patients with brain hydatid cysts. Furthermore, these symptoms could be related to decrease cerebral blood flow secondary to transient anaphylaxis [1].

Computerized tomography and ultrasonography were the main diagnostic methods with 100% and 85% sensitivity in identifying hydatid cyst rupture [2,3].

Urgent treatment should be initiated, relying first on emergency management of anaphylactic shock and later on surgical treatment of the cysts.

CASE REPORT
A 26-year-old man was admitted to the another hospital emergency service with the complaint of abdominal pain accompanied by vomiting and dyspnea. For a while he was unconscious. The pain was not associated with trauma, started suddenly, and was located in the right side of abdomen. He was transferred to our hospital’s emergency department, thinking of epilepsy. At that time he was conscious, only complaining of minor abdominal tenderness. The only physical finding was tenderness in all quadrants. Brain, thorax and abdominal computed tomography (CT) were performed and interpreted as normal except abdominal CT. The abdominal CT examination suggest the perforation of two Echinococcal cyst situated in the subcapsular area of the posterior and anterior lobes of right hepatic. Cyst diameters ranged between 4 to 8 cm. The patient underwent emergency surgery due to intraperitoneal rupture. At that time, he had been in shock; he was rapidly intubated. His blood pressure dropped to 80/50 mmHg, heart rate 135 beats min, respiratory rate 15 bpm. Resuscitation was started with epinephrine (10mcg), methylprednisolone (500 mg). Rapid fluid loading with crystalloid and colloid by central venous cateter were insufficient alone to restore BP and dopamine infusion (6 µg•kg⁻¹•min⁻¹) and 20 mcg epinephrine were administered. The situation progressively improved with continuing fluid loading (total fluid loading:70 ml.kg⁻¹). administering epinephrine and metilprednisolone. Anesthesia was induced by 1.5mg.kg⁻¹ ketamine, 0.5 mcg.kg⁻¹ rocuronium bromide. Sevoflurane 0.25-0.4 % in 50% O2 and 50% air was used for maintenance of anesthesia. Initial blood tests revealed creatinine 1.4 mg/dl, blood glucose 129 mg/dl, sodium 148 mEq/L, potassium 2.9 mEq/L, aspartate aminotransferase 95 IU/L, alanine aminotransferase 108 IU/L, amilase 113 IU/L, creatine kinase 590 IU/L, creatine kinase MB fraction 33 IU/L. Complete blood count revealed leukocytosis (white blood cell 26.700 mm⁻³) with a normal hematocrit and
plateled count. The first arterial blood gas showed metabolic acidosis with wide anion gap (pH 7.20, HCO₃ 6.6 mmol/L, Lactate 45mg/dl, base excess -15.8mmol/L). 80 mEq sodium bicarbonate (8.4% sodium bicarbonate) was infused.

Invasive monitoring with continuous central venous and arterial blood pressure measurements were used. At surgery, two ruptured cysts were seen in segments 7 and 8 of the liver. The patient underwent cystectomy and was transferred to the intensive care unit. He was extubated 7 hours later after the surgery. Two days after admission and previously fully asymptomatic, he dismissed from intensive care unit.

**DISCUSSION**

Hydatid disease, a worldwide zoonosis is usually caused by larval stage of Echinococcus tapeworm. The larval stages of Echinococcus granulosus and Echinococcus multilocularis are involved in humans: cystic echinococcus (CE) (“hydatid disease) and alveoler echinococcus (AE), respectively. The life cycle of Echinococcus granulosus begins in the intestine of a definitive host such as cattle, sheep and rarely humans. The organism penetrates the intestinal wall of the intermediate host and migrates through the host’s circulatory system. It is deposited into an organ forming a cyst. The cycle repeats when the intermediate host is eaten by a new definitive host [1].

Both diseases and parasites have tight links with allergy because of the immunological characteristics that contribute to maintain the larvae in their human host as well as their potential in inducing clinical anaphylactic reactions in some patients. Anaphylactic reactions have systemic signs, including urticaria, edema, respiratory symptoms, and anaphylactic shock due to spontaneous or provoked rupture of the parasitic cyst.

The parasite most commonly locates in the liver (55%-70%), lungs (18%-35%), or uncommon locations such as brain, bones, muscles, adrenals, spleen,extrahepatic bile ducts, thyroid, spine and pelvic region (10%) [2-3].

Hydatid cysts of liver are serious medical problems because of their high rates of recurrence and their life-threatening complications [1-4]. Rupture of hydatid cyst (HC) of the liver may be secondary to trauma or occurs spontaneously. Spontaneous rupture of liver HC into the peritoneum causes an acute abdomen due to peritoneal irritation because of cyst contents. Furthermore, anaphylactic shock may be the first presenting symptom because of systemic reaction of cyst content. Kantarci M and coll. [5] reported a 10 year old boy who was admittted with anaphylaxis due to rupture of hydatid cyst. Cruz I and coll. [6], described a 62 year old woman presented to the emergency department with anaphylaxis. She was operated multivesicular hepatic hydatid cystic disease. On this occasion she presented with a 4-week history of recurrent episodes of transient chest pain, generalized pruritis, flushing and urticaria. Most consensus guidelines for the past 30 years have held that epinephrine is the drug of choice and the first drug that should be administered in acute anaphylaxis.

We preferred to administer intravenous epinephrine and corticosteroid therapy in his initial management. so his condition had improved markedly.

Spontaneous rupture of liver HC caused an acute abdomen. The incidence of peritoneal rupture has been reported to be 5.5% in patients with liver HC [7]. The predisposing factors leading to spontaneous rupture include the increased size of the cyst and increased tension on the cyst wall [8-9]. Superficial localization of the cyst in the liver may also predispose to spontaneous rupture.

Abdominal ultrasonography (US) and CT are the first-line imaging studies in cases of suspected rupture of HC [10-11]. But rupture of the cyst and evacuation of its contents may change the radiological appearance necessitating more sophisticated investigations such as MRI, MRCP, endoscopic retrograde cholangiopancreatography and hepatobilier scintigraphy [12]. Kayihan G and coll. [13] performed that US and CT were the main diagnostic methods with 85% and 100% sensitivity, respectively, in identifying hydatid cyst rupture.In our case brain, torax and abdominal CT scans had been performed and the perforation of two hepatic Echinococcal cysts were visualised clearly.

As a rule, the lungs should be evaluated for associating HC. The reverse is also true for a lung HC. The lung cyst is treated surgically because of effectiveness of antiparasitic drug treatment. In our case we determinated the lungs by this methods.

According to Meyer G Philippe and coll., generalized seizures had never been reported as the initial clinical manifestation of isolated hepatic cyst rupture and initial seizures could be related to decreased cerebral blood flow secondary to transient anaphylaxis at the moment of rupture of the hydatid cyst. They reported a case of hepatic hydatidosis where the first clinical manifestations, generalized seizures after minor head and abdominal trauma [1]. In our patient, loss of consciousness without any
trauma was observed. His brain CT scan was performed thinking of epilepsy at the first.

In conclusion, in endemic areas, the diagnosis must be carefully ruled out in patients experiencing abrupt anaphylactic shock of uncertain etiology. Urgent treatment should be initiated, relying first on emergency management of anaphylactic shock and later on surgical treatment of the cysts.

References
Author Information

Sibel Devrim  
SB Goztepe Education and Research Hospital

Melek Gura  
SB Goztepe Education and Research Hospital

Emine Nursen Koltka  
SB Goztepe Education and Research Hospital

Serdar Yuzer  
SB Goztepe Education and Research Hospital