



# A Rare Cause of Presentation to the Emergency Department with Severe and Refractory Anaphylaxis - Hydatid Cyst Rupture: A Case Report with Literature Review

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## ABSTRACT

Early detection of the etiologic agent is important in patients who present to the emergency department due to anaphylaxis, especially if the anaphylaxis is resistant. Even if there is no history of trauma in resistant anaphylaxis of unknown origin, hydatid cyst rupture should be considered since it is known that the hydatid cyst can rupture spontaneously.

A 15-year-old female patient presented to the emergency department of our hospital with complaints of widespread hot flashes, abdominal pain, and confusion during kickboxing training. Treatment of the patient was started with a preliminary diagnosis of anaphylaxis. Adrenaline infusion was started because of the refractory hypotension and lack of improvement in the clinical findings. The patient's history was taken in more detail because of the resistant anaphylaxis. Abdominal ultrasonography was performed with the suspicion of hydatid cyst rupture due to the patient's history of abdominal trauma while kickboxing. It was observed that the cysts were ruptured in the liver.

This case report emphasizes that hydatid rupture should be among the rare causes of anaphylaxis, especially in regions where hydatid disease is endemic. Ruptured hydatid cyst causing anaphylaxis requires timely diagnosis, treatment, and urgent intervention.

**Keywords:** Anaphylaxis, echinococcus, abdominal pain, cyst rupture

## INTRODUCTION

Infection with *Echinococcus granulosus* (*E. granulosus*) is currently considered an endemic zoonotic disease in the Mediterranean region, including Turkey, due to economic problems (1). It may present with nonspecific complaints such as abdominal pain and jaundice, as well as with anaphylactic reactions due to cyst rupture. We reported this case to emphasize that a very rare disease such as hydatid cyst rupture should be considered among the preliminary diagnoses in patients who present to the emergency services with severe and resistant anaphylaxis

findings but without a history of atopy, asthma, and drug and pollen allergies.

## CASE PRESENTATION

A fifteen-year-old female patient presented to our hospital's emergency department with syncope, generalized flushing, and abdominal pain during kickboxing training. The general condition was moderate and she was conscious and cooperative-oriented. She had generalized flushing. Her arterial blood pressure was 75/45 mmHg, pulse was 110/minute, breath rate was 24/minute, saturation was

97%, and body temperature was 36.7 °C at presentation. The patient was treated with the diagnosis of anaphylaxis. 0.5 mg intramuscular adrenaline was administered. She received a 500 cc isotonic saline infusion in 15 minutes. Ranitidine 1 mg/kg, methylprednisolone 1 mg/kg, and pheniramine 1 mg/kg were administered intravenously. Since the hypotension, flushing, and abdominal pain persisted, 0.5 mg intramuscular adrenaline was administered 2 more times at an interval of 5 minutes. A total volume of 40 cc/kg of isotonic saline was administered. Respiratory distress developed. In addition to 4 liters/minute O<sub>2</sub> supplementation, salbutamol therapy was given. Adrenaline infusion was started for the refractory hypotension and the lack of improvement in clinical findings. Adrenaline infusion was discontinued on the 8th hour of infusion because the clinical findings improved. Generalized flushing, angioedema, and hypotension developed one hour after the discontinuation of adrenaline. The adrenaline infusion was started again. The history was obtained in more detail when the clinical findings did not stabilize and the etiologic agents could not be defined.

The patient did not mention a history of trauma to the abdomen at the first presentation. She had no previous history of hydatid cyst disease. However, she stated that her complaints had started during the training and it was thought that she might have exercise-induced food anaphylaxis. The foods she ate previously were queried in detail. It was learned that she had eaten the same foods she mentioned before the previous training, but no reaction had developed, and the diagnosis of exercise-induced food anaphylaxis was not considered.

The patient's history was taken in more detail because the patient's hypotension was resistant, the urticaria and angioedema were fluctuating, occasional abdominal pain was present, and her clinical condition did not stabilize. It was learned that she practiced kickboxing and experienced trauma to her abdomen. There was no guarding or rebound in the abdominal examination. Considering that the clinical presentation could be explained by the rupture of a hydatid cyst, emergency abdominal ultrasonography (USG) was performed at the 10th hour of hospitalization. As a result of the USG, it was reported that there were ruptured cysts in her liver that had lost pressure, supporting the clinical preliminary diagnosis. Upon the appearance of cysts in the liver, the patient was consulted for surgery and was urgently operated on for cyst resection. After the patient's hemodynamic values remained stable, the adren-

aline infusion was terminated. The Infectious Diseases department was consulted for appropriate antiparasitic therapy. The patient's tryptase level could not be measured due to technical limitations. The patient lived in an apartment in the city and had no history of contact with dogs.

## DISCUSSION

Hydatid cyst is a serious parasitic infection in places where people have close contact with dogs or sheep and is primarily caused by the larvae of *E. granulosus* (2). It can affect all organs, mostly the liver and lungs. While dogs are the main hosts, sheep and herbivores such as cattle and humans are intermediate hosts. Humans are an accidental intermediate host. Humans do not play a role in the biological cycle and the cyst is not transmitted from person to person. Hydatid cysts may remain clinically silent for many years and are often found as incidental findings on ultrasonography performed for unrelated reasons (3).

The most common clinical symptoms of hepatic hydatid cysts are abdominal pain, nausea, and vomiting, especially due to the massive effect of the growing cyst. Other nonspecific symptoms include anorexia, fatigue, and weight loss (4). The disease may be complicated if cyst ruptures occur due to blunt trauma or there are spontaneous increases of intra-cystic pressure (5-8). Allergic reactions such as urticaria, angioneurotic edema, asthma, and anaphylaxis may develop as the antigenic content in the protein structure passes into the systemic circulation as a result of spontaneous or trauma-induced rupture of the cyst.

The frequency of hydatid cyst complications varies between 5% and 40%. Rupture of a liver hydatid cyst to the biliary tree is seen in 6-21% of patients (9). In the literature, there are cases of anaphylaxis and death as a result of the rupture of the cyst during the operation performed for cyst excision. One study examined postmortem results after anaphylactic shock. No ruptured cyst was detected macroscopically, but it was shown that the cysts migrated into the intravascular fluid and caused anaphylaxis (10). In complicated hydatid cyst cases, anaphylaxis carries a high mortality rate of up to 23.5% (5).

In our case, we considered food-related, exercise-induced anaphylaxis based on the patient's first anamnesis. When the history was taken in more detail, we thought of anaphylaxis as a result of hydatid cyst rupture due to the trauma history and resistant anaphylaxis. Santa María

García et al. (11) reported another similar case in a 40-year-old woman with epigastric pain, foreign body sensation in the throat, and shortness of breath after eating pizza, who lost consciousness after 15 minutes with the complaint of unusual anaphylaxis. The reason was that a full laboratory test for an allergic reaction, including a skin prick test, whole blood panel, biochemistry, and basal tryptase levels had normal results. However, Immunoglobulin E (IgE) specific for *E. granulosus* was positive with subsequent imaging confirmation. Although most reported cases of anaphylaxis secondary to hydatid cyst rupture are associated with surgical complications of cyst removal, it is important to note that patients with hepatic hydatidosis may develop anaphylaxis in case of spontaneous cyst rupture or after abdominal trauma (12).

Anaphylaxis is an acute, systemic reaction with a potential risk of death that should be recognized and treated by all physicians (13). Diagnostic criteria have been defined. Regardless of the etiology, cases meeting the diagnostic criteria for anaphylaxis should be treated appropriately.

In the next step, an attempt should be made to determine the etiologic agent. When the etiologic agent cannot be detected in patients who applied to the emergency department due to anaphylaxis, hydatid cyst rupture should be considered even if there is no history of trauma, since it is known that the hydatid cyst can rupture spontaneously.

This case report emphasizes that hydatid rupture should be considered in patients with refractory anaphylaxis, especially in regions where hydatid disease is endemic. Ruptured hydatid cyst causing anaphylaxis requires timely diagnosis and management, and urgent intervention.

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#### Conflict of Interest

The authors declare no conflict of interest.

#### Authorship Contributions

Concept: **Emine Vezir**, Design: **Yuksel Kavas Yildiz, Emine Vezir**, Data collection or processing: **Yuksel Kavas Yildiz, Zeynep Yamcici, Emine Vezir**, Analysis or Interpretation: **Yuksel Kavas Yildiz, Emine Vezir**, Literature search: **Yuksel Kavas Yildiz, Emine Vezir**, Writing: **Yuksel Kavas Yildiz, Emine Vezir**, Approval: **Yuksel Kavas Yildiz, Zeynep Yamcici, Emine Vezir**.

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