

## Anaphylactic shock as the only clinical manifestation of hepatic hydatid disease

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### Abstract

The case of a 35-year-old man is reported who developed a spontaneous anaphylactic shock as the only clinical manifestation of hepatic hydatidosis. Dermatologists should consider asymptomatic hydatid disease in the differential diagnosis of anaphylactic reactions, particularly in patients from regions in which echinococcosis is endemic.

### Introduction

Hydatid disease is a worldwide parasitic infection caused by adult or larval stages of cestodes who belong to the genus *Echinococcus*. From the epidemiologic and clinical point of view, *E. granulosus* and *E. multilocularis* are the most important species, as they cause cystic and alveolar echinococcosis, respectively.<sup>1</sup> We present a patient who developed an anaphylactic shock as the only clinical manifestation of hepatic hydatid disease.

### Case Report

A 35-year-old Caucasian man was admitted to the Department of Dermatology, University of Patras, Greece, with the sudden onset of generalized urticaria, pruritus, rhinal congestion, dyspnea, dizziness, unproductive cough, vomiting, and discomfort. On physical examination, he was found to have a generalized urticaria, cyanotic lips, a positive shock index (blood pressure, 90/60 mmHg; cardiac rate, 130 beats/min), and a respiratory rate of 36/min. Lung examination showed expiratory wheezing, whereas no obvious tenderness or organomegaly was found on abdominal examination. Apart from a sinus tachycardia, the electrocardiogram (ECG) was unremarkable. An anaphylactic shock was diagnosed and the patient's condition improved rapidly subsequent to intravenous administration of steroids, H<sub>1</sub> and H<sub>2</sub> antagonists, and fluid. A similar episode occurred on his second hospitalization day. He had no history of abdominal trauma, allergy, systemic mastocytosis, hereditary angioedema,

pheochromocytoma, pancreatic tumor, carcinoid, jaundice, diabetes, hypertension, or coronary heart disease, and there was no suggestion of any cause for his anaphylactic episodes, such as drugs, food, exposure to chemical or physical agents, or insect bite. Apart from a leukocytosis (16,000/mm<sup>3</sup>) without eosinophilia and an increase in serum levels of glutamic pyruvic transaminase (SGPT) (79 U/L; normal range, 5–40 U/L),  $\gamma$ -glutamyl transpeptidase (GGT) (57 U/L; normal range, 10–50 U/L), and immunoglobulin E (IgE) (244 IU/mL; normal range, 10–180 IU/mL), the results of routinely performed hematologic and biochemical investigations were within normal limits. Serologic investigations for infectious diseases and a series of allergic tests for drugs and foods revealed negative results.

Chest X-ray showed an elevation of the right hemidiaphragm, but no pathologic findings in the lungs and mediastinum. Abdominal ultrasonography and computed tomography (Fig. 1) revealed two cystic lesions in the lower right lobe of the liver. These findings were considered to be consistent with the diagnosis of hepatic hydatid cysts and the patient was transferred to the Department of Surgery for surgical treatment. When the peritoneum was opened, a partially calcified 7.8-cm hydatid cyst and a small 2-cm satellite cyst were visualized at segment V and at the dorsal aspect of the liver at segment VI adhering to the gallbladder. After controlled evacuation of the content of the cysts, three minute fissures were detected at the bottom of the large cyst. A wide capsectomy of the cysts and simultaneous resection of the gallbladder were performed. By suturing the edges of the resected wall of the cysts and the hepatic parenchyma with running interlocking



**Figure 1** Computed tomography of the hepatic hydatid cysts

sutures, hemostasis and obliteration of the small biliary ducts were achieved.<sup>2,3</sup> The patient made an excellent postoperative recovery without any complications. Anthelmintic treatment (albendazole 10 mg/kg/day) was given postoperatively for four cycles of 1 month each. No recurrence of either the hydatid disease or the anaphylactic reactions was observed during a 5-month follow-up of the patient.

## Discussion

Cystic echinococcosis is endemic in Mediterranean countries, the Middle East, Australia, South America, south-eastern Africa, and in some states of the USA. It is characterized by high morbidity, considerable mortality, and a negative socio-economic impact.<sup>4,5</sup> Humans become infected accidentally as intermediary hosts and house the larval form of the parasite transmitted through food-borne exposure to the feces of carnivores containing eggs of *E. granulosus*.<sup>6</sup> The most commonly affected organs are the liver (60%) and the lung (40%).<sup>7,8</sup> Hepatic hydatid cysts consist of an inner layer developing from the parasite and an external layer derived from the liver.<sup>6</sup> In most patients, they are clinically inapparent for a long period of time and can be detected incidentally in asymptomatic patients undergoing abdominal ultrasonography for unrelated reasons.<sup>9</sup> Symptomatology, when present, depends on the anatomic location, size, and phase of development of the hepatic cysts.<sup>10</sup>

Cutaneous manifestations of hydatid disease are either specific (skin hydatidosis and hepatic-cutaneous fistulae) or nonspecific (pruritus, urticaria, generalized pustulosis, and anaphylactic reactions).<sup>11–15</sup> Spilling of the highly antigenic hydatid cyst fluid,<sup>16</sup> due to accidental surgical or post-traumatic rupture, can trigger severe anaphylactic reactions and shock.<sup>17–19</sup> Spontaneous anaphylactic phenomena in asymptomatic patients with hydatid disease, however, are extremely rare, and only a few cases have been reported in which an anaphylactic

reaction was the initial manifestation of hydatidosis.<sup>20–25</sup> In our case, leakage of the cyst through the observed three fissures might have facilitated the contamination of the bloodstream with the antigenic components of the cystic fluid, resulting in a hypersensitivity reaction, probably mediated by IgE.

In conclusion, particularly in patients from regions in which echinococcosis is endemic, dermatologists should consider possible leakage of an asymptomatic hydatid cyst in the differential diagnosis of severe anaphylactic reactions and shock.

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