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Case Report

Latent Fatality Due to Hydatid Cyst Rupture after a Severe Cough Episode

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Erdogmus, B., Yazici, B., Akcan, Y., Ozdere, B.A., Korkmaz, U. and Alcelik, A. *Latent Fatality Due to Hydatid Cyst Rupture after a Severe Cough Episode*. Tohoku J. Exp. Med., 2005, **205** (3), 293-296 —— Hydatid disease is a parasitic disease caused by *Ecinococcus granulosus* characterised by cyst formation in various organs. The infestation mostly involves the liver. Hydatid cysts of the liver can rupture either spontaneously or due to trauma. Incidence of rupture is about 3-17% of all cases with hydatid disease. Unless treated surgically, rupture can result in death. Here, we present a case of hydatid cyst ruptured after a severe cough episode and disseminated first to the subcapsular area, then to the peritoneal space. Probably due to a decrease in parenchymal pressure in the liver after decompressive effect of rupture, the patient felt an improvement in abdominal pain, refused operation, and left the hospital on his own responsibility. This unfortunate relief resulted in a delay of 55 hours in management. The leakage of liquid materials into peritoneal space resulted in a severe inflammatory reaction and eventually death of the patient. The patient died of a late peritonitis rather than anaphylaxis, which is the most common reason for death in such patients. As a conclusion, physicians should be aware of a temporary relief in abdominal pain after cyst rupture that may cause a delay in management and in turn loss of patient due to peritonitis. ———— cough; hydatid cyst; imaging; rupture

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Hydatid disease is a parasitic infestation caused by the larval stage of *Ecinococcus granulosus*. Hydatid disease is known to be endemic in Middle Eastern, Mediterranean regions, and Australia (Camunez et al. 1986). Hepatic hydatid cyst can rupture either spontaneously or due to trauma (Bedirli et al. 2002). The incidence of rupture is about 3-17% of all patients with hydatid cyst. It can cause fatal results if the patient is not operated immediately after the time of rupture (Kumar et al. 2002). Pulmonary hydatid cysts can erode the surrounding parenchyma and perforate into bronchial system or pleura upon cough, effort or trauma or spontaneously (Akhan et al. 1994). To the best of our knowledge, a case of death due to peritonitis after hepatic hydatid cyst rupture caused by cough has not been reported in the literature.

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Case Report

Fifty-six year-old male patient experienced a sudden right upper quadrant pain following a vigorous cough episode. About 5 hours later, he had another cough episode, which ameliorated his abdominal pain, and the patient felt comfortable to some degree. Despite this partial relief, the patient came to our emergency service. He had no pruritus, erythema, local or generalized edema, stridor or dyspnea. No history of blunt trauma was present in his clinical history. He has been under control for seven years because of chronic obstructive pulmonary disease. In the physical examination of the patient, right upper quadrant tenderness and hepatomegaly were present. The laboratory investigations revealed leukocytosis, elevated alkaline phosphatase, and normal bilirubin levels. In chest radiography, bilateral chronic fibrotic and emphysematous changes were detected. Abdominal ultrasonography (US) showed a cystic lesion (10 × 15 cm) with irregular borders, internal echoes and septa in the right lobe of liver. The superolateral border of the cyst was incomplete and in the subcapsular area, there was a fluid collection with internal echoes (Fig. 1). The biliary ducts were in normal calibration. In computed tomography (CT), there was a hypodense lesion in cystic density with thin and lobulated borders, which extended subcapsullary (Fig. 2). In fast spin echo (FSE) T2-weighted axial magnetic resonance imaging (MRI) sequences, it was noted that hypointense ruptured membrane and subcapsular fluid collection were in connection with the cystic lesion in the liver parenchyma (Figs. 3A and 3B). With these findings, diagnosis of subcapsular, ruptured hydatid cyst was made. Despite all warnings and recommendations, the patient and his relatives did not accept operation. Upon his general condition deteriorated, he again came to our hospital after 55 hours. In physical examination, acute abdomen was detected and the patient has died during preparation for surgical operation. In autopsy, purulent fluid and edematous intestinal segments were found in the abdomen. The cause of death of the patient was defined as peritonitis developing secondary to hydatid cyst rupture.

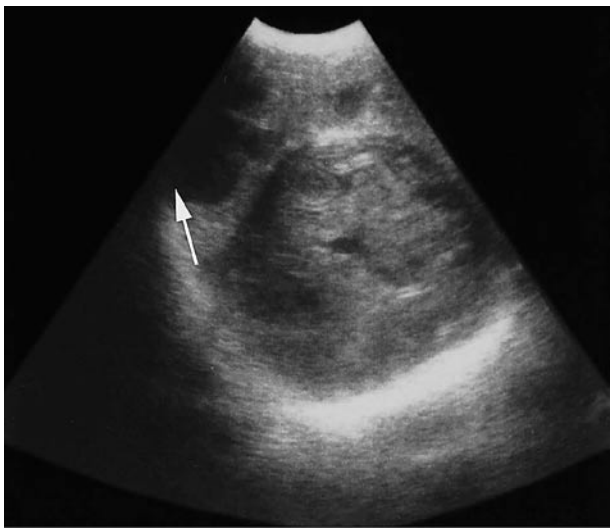


Fig. 1. Irregularly marginated cystic lesion. The ultrasonographic examination shows an irregularly marginated cystic lesion with internal echogenic septa in the right lobe of the liver. There was an irregularity in the superolateral wall of the cyst and subcapsular fluid collection (white arrow).



Fig. 2. The lobulated cystic lesion. The CT imaging shows a lobulated, thin walled, hypodense lesion which was extending to the subcapsular area (white arrows).

Discussion

The hydatid disease affects most commonly the liver, lungs, spleen, and kidney and occasionally the brain and muscle tissue (Camunez et al.

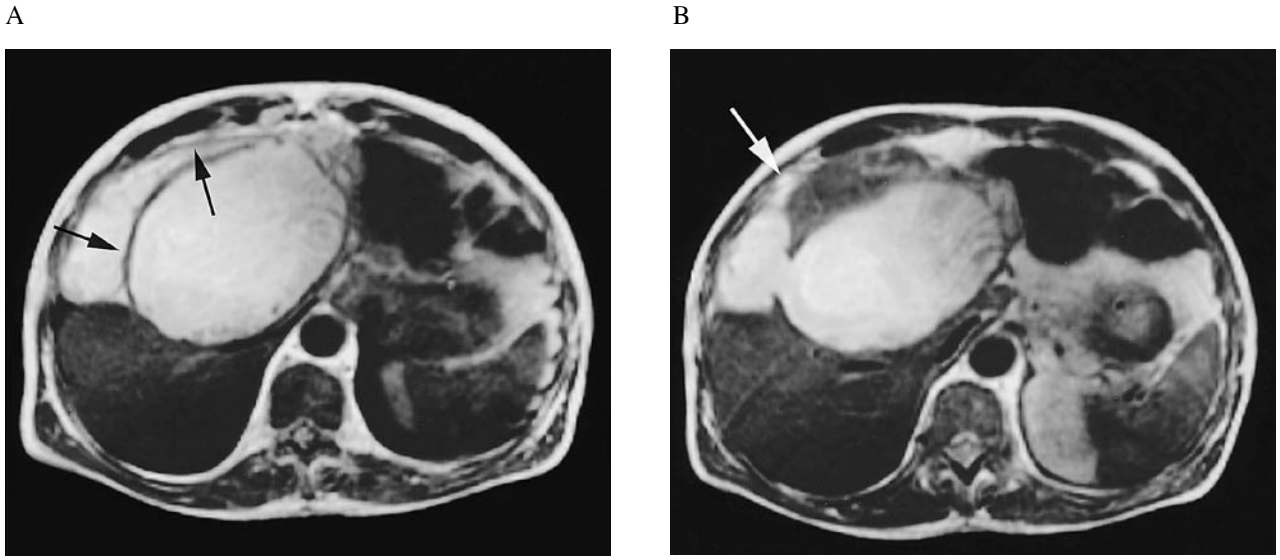


Fig. 3. The ruptured cystic lesion. FSE T2-weighted axial of MRI sequence shows hypointense ruptured membrane in A (black arrows) and subcapsular fluid collection in B (white arrow).

1986; Kiliç et al. 1997). Unless it causes complications, the disease might continue to progress without symptoms. Hydatid cysts can rupture either spontaneously or as a result of trauma over degenerated membranes of the cyst (Camunez et al. 1986). Probably, the rupture in our case was due to the degenerated cyst because of increased intraabdominal pressure caused by severe cough episode.

Generally, the first imaging modality to be used is US. In ultrasound examination of ruptured cases, regression in the size of the cyst, freely ondulating membrane inside the cyst, echogenic hydatid material in the biliary ducts which has acoustic shadow can be seen in US. (Barki and Charuzi 1985; Lewal and Mccorkell 1986). In CT, hydatid cyst has a wall which enhances after contrast administration, internal septations, and undulating membranes in the cyst and calcifications in the wall of the cyst. The point, where the irregularity of the wall is seen, shows the probable site of rupture (Martti-Bonmati et al. 1988; Kumar et al. 2002). MRI is commonly used in the cases, when the definite diagnosis was not obtained with US and CT. Hypointense layer that is seen in the wall of cyst, differentiate hydatid cyst from the other cysts. The interruption of the hypointense layer of the cyst wall is the direct finding of the rupture. The air-fluid levels and changes in the signal intensities inside the cyst are indirect findings (Mendez Montero et al. 1996). In our case, the definitive diagnosis was made by the irregularity in the cyst wall which was hypointense in T2 weighted sequences.

In the complicated cases, the lesions located in the liver can perforate into biliary system, pleura, pericardium, or peritoneum (Camunez et al. 1986). They can rupture via three mechanisms. In the first type, the endocyst is ruptured but the pericyst is intact. Hydatid fluid enters the space between the endocyst and pericyst and the endocyst collapse. In CT and US, the membrane is undulated without a change in the size of the cyst. In this rupture type, the development of an infectious or allergical condition is impossible. In the second type of rupture, both pericyst and endocyst are ruptured. The content of the cyst disseminate into the bronchial system or biliary ducts as the pericyst combine with these structures. In the third type, endocyst and pericyst rupture and the content of the cyst directly disseminate into peritoneal or pleural cavity (Lewal et al. 1986). If a delay occurs in the treatment, effusion and empyema may develop in the cases of pleural dissemination. Similarly, peritonitis and peritoneal hydatidosis occur in the cases of peritoneal dis-

semination. As a result of direct dissemination of hydatid cyst to serous spaces, the cyst contents can cause allergy and anaphylaxis (Camunez et al. 1986). When our case was admitted to our emergency service after a severe cough episode, the second type rupture was present. After recurrent cough episodes, the third type rupture occurred, and cyst contents disseminated into the peritoneal space, causing latent peritonitis.

The main clinical manifestations of anaphylaxis due to hydatid cyst, which develop within minutes of the rupture, are high fever, pruritus, edema of the lips and eyelids, dyspnea, stridor and rhinorrhea (Bitton et al. 1992). None of them was present in our patient on either initial or second administration. We have concluded that peritonitis and shock secondary to severe inflammatory response over peritoneal surfaces are responsible for the cause of death in our patient, although anaphylaxis is the most frequent cause of death in cases of hydatid cyst rupture. This is the second case in literature reporting a fatality secondary to a late peritonitis after hydatid cyst rupture rather than anaphylaxis (Ivanis et al. 2003). The differentiating feature in our patient is that he experienced cyst rupture after a severe cough episode. The common event in both cases was the patient's refusal of appropriate and timely surgical intervention. We believe that in cases not experiencing a quick anaphylactic reaction, the pressure decrease in liver parenchyma after spillage of cyst contents into peritoneum might cause a temporary relief. This false relief in turn would cause the patient and his relatives to give a wrong medical decision as to reject surgery. The physicians should be aware of this temporary relief after spontaneous cyst decompression and warn the patient for the imminent poor prognosis unless treated surgically.

As a result, the degenerated wall of the cyst can rupture due to a severe cough episode. The ruptured hydatid cyst needs emergent surgical intervention. Otherwise, fatal results may occur in untreated cases. The death is caused mostly by anaphylaxis or in rare cases by a late peritonitis.

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