

Peritoneal Hydatid Cyst mimicking Peritoneal Seeding; a case report and review of literature

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Introduction

Hydatid cyst is caused by infection with a parasite belonging to the *Echinococcus* family. Humans are the intermediate host of this disease and get infected directly or indirectly after the parasite eggs enter the digestive system. According to the WHO, it is considered a neglected disease world-wide, as it still remains highly prevalent despite the improvement of public health and following sanitary protocols by humans (1,2). This disease is mostly asymptomatic and is incidentally detected (3). Usually, only one cyst is formed during infection, and the formation of several cysts and the involvement of several organs is not common. The most commonly involved organ is the liver, followed by the lung. The involvement of other organs is much less common, and the involvement of the peritoneum and omentum is very rare, and is usually caused by abdominal trauma, the rupture of a cyst in the liver or other organs, and the release of parasites leading to the contamination of the abdominal space (4). In this study, we introduce a case of hydatid cysts in liver, spleen and peritoneum, in which the radiological appearance and clinical symptoms had raised high suspicion of abdominal malignancy and peritoneal seeding.

Case History/examination

64-year-old male farmer with elementary education, living in one of the villages of Zanjan, one of the central provinces of Iran, visited the specialized surgery clinic of our center 9 months ago with a complaint of vague abdominal pain dominantly in the epigastric area. He had lost more than 10 kg of weight during this period, and in the last month, he mentioned symptoms such as loss of appetite, weakness, and lightheadedness. The pain was continuous and was not related to eating, physical activity or physical position. The patient did not mention nausea and bowel pattern was normal. The patient had been under treatment with metformin 1 gram daily for the past 5 years due to diabetes and had no history of other disease or medication. The patient had no history of surgery, alcohol use, smoking, or opium consumption, and did not mention any medication or food allergy. The patient's brother had a history of colorectal cancer and there was no history of any other significant diseases in the first degree family members. On physical examination, vital signs and general appearance were normal. Abdominal exam did not find anything abnormal. Also, general physical exam did not contain any pathological findings.

Methods

The patient was admitted with initial suspicion of gastrointestinal malignancy and underwent diagnostic and therapeutic measures. In the preliminary tests, there were no other abnormal findings except for low hemoglobin, MCV, serum iron and ferritin, and high FBS and HbA1C. Abdominal ultrasound showed evidence of a cystic liver lesion, splenomegaly and multiple spleen lesions favoring a hydatid cyst. Next,

the patient was tested for hydatid serology, which was negative. For further investigation, a CT scan was performed for the patient. CT scan result of the lung was not remarkable. In the CT scan of the abdomen and pelvis, there was evidence of splenomegaly (AP: 174mm) and the appearance of at least 6 cystic lesions (with the largest being 92x108mm), one calcified cyst (23x33x43mm) in the 6th segment of the liver. In addition, there were a large number of peritoneal nodules in different regions of the abdomen (with the largest being 48x27mm) and at least 6 hypodense structures (with the largest being 45x67x54mm) in the middle part of the omentum and above the umbilicus, for which hydatid cysts, primary neoplastic or metastatic lesions to the peritoneum were included in the differential diagnosis (fig. 1). Considering the anemia and weight loss and the possibility of peritoneal seeding, hydatid cyst could not fully explain the condition, so the patient underwent further diagnostic measures to investigate the cause of anemia. In colonoscopy, the only abnormal finding was internal hemorrhoid grade I, and endoscopy had no other significant findings except for small size sliding hiatal hernia.

Later, patient underwent diagnostic laparoscopy. One port was placed below the navel and the other was placed on the right side. In the general exploration of the abdomen, there was evidence of extensive involvement of the omentum with many nodular lesions, splenomegaly, and several large cysts of the spleen and one liver cyst. Stomach, small intestine and colon were examined completely and had no pathological findings. Several samples of nodular omentum lesions were obtained and sent for frozen section examination. The initial pathology report showed omental hydatid lesions. Therefore, the patient underwent laparotomy and splenectomy. Macroscopically, the size of the spleen was 14 x 16 x 19 cm and its weight was 686 grams with an irregular surface containing several cysts (fig. 2). Also, parts of the omentum that had evidence of involvement with hydatid lesions were removed (fig. 3). Abdominal cavity and pelvis were thoroughly re-examined, no other pathology was found and surgery was complete.

Later, patient received 13-valent pneumococcal vaccine and was treated with albendazole 800 mg daily, oral iron and folic acid, and was discharged 3 days later in favorable general condition. Patient was also advised to receive the 23-valent pneumococcal vaccine two weeks later. The final pathology report also showed omental and splenic hydatid cyst (fig. 4). In 3 and 6 months of follow-up, the patient's abdominal pain had improved and general examinations were normal. Liver enzymes had not increased significantly. A control ultrasound was also performed and there were no other pathological findings except for calcified hepatic cyst.

Conclusion and Results

According to the present study, hydatid cysts of the peritoneum and omentum, despite considerably low prevalence, may occur in association with hydatid cysts in other regions of the body, including the liver and spleen. Although these cysts are usually a result of abdominal trauma or hydatid cyst surgery, they can also be caused by hematologic or lymphatic dissemination or by rupture of the cyst adventitia. CT scan still remains the best diagnostic method for hydatid cyst, but the appearance of peritoneal and omental hydatid cyst may be mistaken for malignant lesions and peritoneal seeding. This case is significant as misdiagnosis may delay treatment and lead to risks of rupture, peritonitis and anaphylactic shock and imminent death. Surgery and removal of the cyst along with anthelmintic treatment is the best method of treatment in these cases and can prevent the aforementioned complications and improve the survival of the patients.

Discussion

Tapeworms from the *Echinococcus* family have been introduced since the 1950s as a pathogenic parasite for humans. Two important species of this parasite include *E. granulosus* and *E. multilocularis*, which cause cystic echinococcosis (CE) and alveolar echinococcosis (AE), respectively, among which infection with CE is more common. The mature worm exists in the small intestine of carnivores, and the eggs are released via the animal's feces and contaminate water and food. Food materials contaminated with *Echinococcus* eggs are swallowed by intermediate hosts, including herbivores, and the eggs enter the human body directly (contact with infected animals) or indirectly (eating contaminated food or water) and the larvae spread through the human body hematologically and via the lymphatic route (5,6).

This parasite is distributed all over the world, but the endemic areas include Oceania, China and the central

regions of Asia, central and southern Russia, Middle East, Mediterranean regions, parts of Africa and North America (6). CE is considered an important parasitic disease in the world, and is an important concern for developing and low-income countries due to significant mortality and morbidity. According to the WHO report, this disease is one of the 17 important neglected parasitic diseases. More than one million people are infected annually and about 20 thousand people die due to the disease. Disability-adjusted life-years (DALYs) are estimated to be more than 800,000 years and the annual treatment cost is over 3 billion dollars (6,7). This disease usually occurs in the 40s and 50s and is slightly more common in women than men, and living in urban areas and contact with dogs and livestock bears a greater chance of infection (3). CE is usually asymptomatic and is usually found incidentally in unrelated examinations. Although in many cases, several diagnostic measures are used, CT scan is the gold standard for hydatid cyst diagnosis (8). Serological tests are only used to confirm the diagnosis, and negative test results do not rule out CE. It may take years from the time the parasite enters the human body until the symptoms develop, and the development of symptoms depends on the size of the cyst and its anatomical location (9). If symptoms develop, dull abdominal pain is the most common symptom. Usually, during CE disease, only one cyst is formed and multi-organ involvement is not common. The most common site for cyst formation is the liver, followed by the lungs. Involvement of the CNS, muscles, and spleen rarely occurs, and involvement of the peritoneum and omentum is extremely rare (3).

The presented patient was an elderly male living in the village with occasional contact with livestock, who presented with a complaint of vague abdominal pain and weight loss, as well as a history of colorectal cancer in the first degree family. Due to iron deficiency anemia and presence of red flag symptoms, preliminary investigations aimed to rule out malignancy. Although cystic lesions of the liver and spleen favoring hydatid disease were found in ultrasound, serology testing of hydatid cyst was negative. In addition to confirming the presence of a calcified liver cyst, the CT scan showed an enlarged spleen containing several cystic structures and evidence of extensive involvement of the omentum and peritoneum, which looked similar to metastatic lesions and peritoneal seeding. Therefore, we decided to perform a diagnostic laparoscopy for this patient, the result of which was the involvement of the peritoneum and omentum with hydatid disease.

In recent studies, patients with CE mostly had one cyst, and in a study conducted in 2020 on 501 CE patients over 15 years, the prevalence of peritoneal involvement was only 1% (3). Study results have shown that peritoneal hydatid cyst usually occurs secondary to trauma or abdominal surgery and due to the rupture of a liver cyst in the abdominal cavity. Despite this, the presented case had no history of abdominal surgery or blunt trauma to the abdomen. There are various hypotheses regarding the development of primary hydatid cyst in the peritoneal cavity, none of which have been proven. Although, it has been suggested that the localization of hydatid cyst in the peritoneum can be a result of the migration of the liver cyst through the blood and lymphatic circulation or the rupture of the adventitia without rupture of the cyst membrane (4).

The main approaches of treatment are medical treatment with anthelmintic drugs (usually Albendazole 10 mg/kg), surgery or both. The aim of treatment is to eliminate the cyst, minimize complications and prevent recurrence. In this regard, surgery is still the best therapeutic approach and will convey the best results if combined with medical treatment. Medical treatment is effective in stopping parasitic growth and is usually not enough alone. Isolated medical treatment is indicated when the patient is not a suitable candidate for surgery (10). In the patient in question, due to splenomegaly and the involvement of all parts of the spleen, splenectomy was performed, and in relation to the peritoneal and omental cysts, due to the high risk of perforation leading to peritonitis and anaphylactic shock, all parts involved with cysts were removed. Also, due to the calcification of the liver cyst, this cyst remained intact and the patient was prescribed oral Albendazole for 3 months.

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