

Isolated Peritoneal, Mesenteric, and Omental Hydatid Cyst: A Clinicopathologic Narrative Review

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Abstract

Hydatid disease (HD) is caused by *Echinococcus granulosus* and is endemic in many parts of the world. This parasitic tapeworm can produce cysts in almost every organ of the body, with the liver and lung being the most frequently targeted organs. Peritoneum, omentum, and mesentery are among these unusual locations, which can cause diagnostic challenge and treatment delay. This review provides information on the reported cases of the peritoneal, omental, and mesenteric hydatid cyst in the world during the last 20 years. During the last 20 years, there have been 49 published cases of hydatid cysts in the peritoneum, mesentery, and omentum. Among the reported cases in the English literature, the most common presenting symptom has been chronic abdominal pain and the method of primary diagnosis has been ELISA and ultrasonography. The best treatment modalities have been surgical excision, with and without adjuvant therapy, with albendazole and scolicidal agents. The published follow-up studies showed a low recurrence rate.

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Introduction

Echinococcus granulosus is a tapeworm that causes hydatid disease. Hydatid disease (HD) as a parasitic disease is caused by the larval stage of *Echinococcus granulosus*.¹ HD is endemic in the Middle East, Africa, South America, New Zealand, Australia, Turkey, Iran, India, and Southern Europe, but concentrations are common in almost every part of the world.² This parasite lives in the intestine of dogs, wild canines and carnivorous animals (definitive host). It can also involve both domestic and wild animals. Humans become the accidental intermediate hosts by ingesting *Taenia* eggs, which causes slow growing hydatid cysts containing many thousands of protoscolices that form daughter cysts. The liver is the most commonly involved organ by the echinococcal cysts (65-70%) followed by the lungs (25%).³ However, this parasitic tapeworm can produce cysts in almost every organ of the body. In about 8-10% of the cases, the cyst tends to appear in unusual body sites in various geographical areas of the world.⁴ Unusual sites of this disease such as breast, adrenal, appendix, peritoneum, omentum, and mesentery⁴⁻⁷ can frequently cause diagnostic problems, which can give rise to an increased risk of diagnostic delay and many potentially serious complications.¹

What's Known

- Hydatid disease is common in countries such as Iran.
- It can occur in unusual locations such as Iran.

What's New

- Little is known about the general clinical and paraclinical diagnosis of the rare locations of hydatid disease in the peritoneum, omentum, and mesentery.

The incidence of HD involving the peritoneal cavity and related organs (omentum and mesentery) is about 2%. This benign disease can cause substantial morbidity and mortality in these organs.² As Iran is an endemic country for HD, in any unusual presentation and location, this disease should be considered in patients presenting with cysts or cystic masses in every organ of the body. However, we did not find any comprehensive review regarding the peritoneal, omental, and mesenteric location of this disease.

The aim of this study was to review the English literature in the past 20 years concerning the rare occurrence of peritoneal, omental, and mesenteric hydatid cyst and its clinicopathologic findings. Essential findings such as demographic findings, presenting symptoms, method of primary diagnosis, laboratory findings, operative findings, treatment modalities, and follow-up studies were separately included in each case.

During the last 20 years, there have been 49 reported cases of peritoneal, omental, and mesenteric hydatid cysts in the English literature. Table 1 shows the number of cases from different parts of the world. Most common geographic locations are in Asia (India) and sheep-raising countries of the Mediterranean such as Iran, Turkey, and Greece.

Demographic Findings

Table 2 shows the most important and common findings of hydatid cysts in the above-mentioned geographical locations. The 49 reported patients in the English literature with peritoneal, omental, and mesenteric hydatid cysts were 27 women and 22 men, aged 2-85 years. Most of the cysts were large between 5 cm to 45 cm.

Sign and Symptoms

All except one patient presented with abdominal pain and distension. There were 1 case with painless abdominal distension,²⁹ 3 (6.1%) cases of vomiting, 3 (6.1%) cases of nausea, 3 (6.1%) cases of weight loss, 3 (6.1%) cases of anorexia, and 3 (6.1%) cases of fever.^{8-11,41,46} Unusual symptoms such as irregular vaginal bleeding were also reported in individual cases.¹² There were 7 (14.2%) patients with incidental finding of abdominal or appendiceal mass, which turned out to be hydatid cyst of the peritoneum.^{8,13,29,30,35,41,42} Two patients underwent emergency operation because of abdominal tenderness, muscle guarding, and clinical impression of peritonitis.^{10,36}

Diagnostic Methods

Preoperative diagnosis was made by different methods, namely:

Table 1: Countries and the reported number of peritoneal, omental, and mesenteric hydatid cyst cases

Country	Number
India	24 ^{2,8-28}
Turkey	11 ²⁹⁻³⁴
Iran	5 ³⁵⁻³⁹
Greece	3 ⁴⁰
Morocco	2 ^{41,42}
Italy	1 ⁴³
Tunisia	1 ⁴⁴
USA (immigrant from Peru)	1 ⁴⁵
Pakistan	1 ⁴⁶
Total	49

Table 2: Most common clinicopathologic characteristics of hydatid cysts of the abdominal cavity (omental, mesenteric, and peritoneal)

Most common country	India (24 cases, 48.9%)
Age	2-82 years (42±26)
Sex (F/M)	27/22
Most common presenting symptom	Abdominal pain and distension
Duration of disease	5 days to 2 years (180±273 days)
Anemia	12.2% (6 cases)
Eosinophilia	10.2% (5 cases)
Positive ELISA test (for hydatid cyst) in 9 performed cases	67% (6 cases)
Most common preliminary diagnosis	Mesenteric cyst, ovarian cyst
Most accurate diagnostic method	Imaging (Ultrasonography and CT-scan)
Most common medical treatment	Pre- and post-treatment albendazole
Most common surgical treatment	Excision

- Laboratory findings: There were no specific and consistent laboratory findings in these patients. However, anemia and increased eosinophil count were reported in 6 (12.2%) and 5 (10.2%) cases, respectively.^{8,11,12,14,46}
- Enzyme-linked immunosorbent assay (ELISA): This method was performed in only 9 cases, of which 6 (67%) were positive with titers between 1/80 to 1/160.^{8,14-17,40,41,45,46}
- Casoni test: This test was reported in two cases with negative results.⁴⁰
- Imaging studies: CT-scan and ultrasonography have been reported as either a well-defined large and huge heterogeneous anechoic or hypoechoic multiseptated cyst or multiple small cysts with a honeycomb pattern and spoke-wheel appearance with or without calcification.⁴⁷ Ultrasonography was reported in all cases, however, to a lesser extent CT-scan was

also performed in cases of peritoneal HD.²⁹

- Pathologic findings: Main histopathologic finding in HD of the peritoneum, omentum, and mesentery is a cyst. The cyst wall contains an outer fibrous laminated layer and an inner germinative layer. There are scolices and hooklets within the cyst lumen. The surrounding tissue shows infiltration of chronic inflammatory cells, including lymphocytes and eosinophils.⁴⁸

Treatment

The most common successful treatment with no recurrence has been surgical excision of the cyst with postoperative antiparasitic medical treatment (albendazole). In cases with the preoperative diagnosis of hydatid cyst, preoperative albendazole has been used, which can be helpful in preventing dissemination of the scolices during surgery and also preventing recurrence as it can sterilize the cyst and decrease the tension in the cyst wall.^{9,14,15,18-21,31} Intraoperative hypertonic saline or saline nitrate solution can also kill daughter cysts to prevent further spread and anaphylactic reaction.^{40,49}

Discussion

HD is a common parasitic infestation in sheep and goat farming countries such as Iran, Turkey, and India. Additionally, it has also been reported in North America and a few European countries such as Greece and Spain. Consequently, this disease is one of the main zoonoses in both developed and developing countries.⁵⁰ According to epidemiological studies, this disease has been reported from all continents except for the Antarctica. Clearly, many people are involved with HD worldwide.⁵¹

The cysts of *Echinococcus granulosus* can be seen in every organ, even in unusual sites such as peritoneum, omentum, and mesentery of the bowel.⁴ Most cases of HD in the above-mentioned locations have been accompanied by the lung and liver hydatid cysts or secondary to the ruptured hepatic liver cysts.^{1,52} It means that the primary location of HD in the peritoneum or involving omentum and mesentery of the bowel is an extremely rare occurrence.¹⁶ However, it should be considered as an important differential diagnosis in endemic countries.

To the best of our knowledge, 49 cases have been reported in the English literature during the last 20 years. As shown in table 1, most of the reported cases were from India, Turkey, and Iran. These patients remain asymptomatic for a long time and mostly present secondary to large size and pressure effect.¹⁶ Therefore, the

most common symptoms have been chronic abdominal pain, presence of fever, weight loss, and anorexia. Other accompanying signs such as abdominal tenderness are not common.⁸ Moreover, the presence of palpable abdominal mass in these locations of HD is very rare.²⁹

Based on experience, laboratory findings such as marked eosinophilia (if available) can be helpful in preoperative diagnosis.⁸ However, no increase in eosinophil count has been reported in more than 90% of the past cases.¹⁰ In patients with primary involvement of peritoneum, omentum, and mesentery by HD, liver function test remains normal.¹⁶

Serological tests such as immunoelectrophoresis and ELISA have been reported for the diagnosis, screening, and postoperative follow-up of HD recurrence. Among these, ELISA is the most commonly used method. The Casoni test was also used for the diagnosis of such cases; however, it is no longer practiced due to low sensitivity.^{32,40} The sensitivity and specificity of ELISA test is high in the liver and lung but is much lower in unusual locations (25-65%) in the HD of other organs. This is because the capsule isolates the parasite from the host's immune system that yields into a negative test.² In the majority of intra-abdominal peritoneal, omental, and mesenteric HD cases, ELISA test for *Echinococcus granulosus* has not been performed before surgery. Such scenario is only reported in 9 cases, of which 6 (67%) were positive.^{8,14-17,40,41,45,46} Serological tests for diagnosing hydatid is valuable only if it is positive.⁴²

Imaging investigations have an important role in the preoperative diagnosis of HD cases. The preferred imaging method depends on the involved organ and the growth phase of the hydatid cyst.²⁹ Conventional plain abdominal X-ray shows an eggshell calcification.¹⁶ Based on experience and according to the case reports on peritoneal, mesenteric, and omental hydatid cysts, ultrasonography of the abdomen is the most commonly performed first-line imaging modality to identify the cause of abdominal pain, distension, or mass.⁴⁷ It has an approximate sensitivity of 90-95%. Most often, ultrasonography shows a large solitary unilocular lesion or multiple anechoic or hypoechoic, well-defined cysts with or without daughter cysts, and imaging characteristics of internal septations.^{37,53} Hydatid sands may be visible by shifting patient's position during the procedure. When the fluid pressure in the cyst rises, the inner membrane will be detached which produces snake/serpent sign. The collapse of inner membrane produces Water-Lily sign.¹⁶

CT-scan is another helpful modality for the diagnosis of intra-abdominal extraintestinal HD, as it permits imaging of the entire abdomen.¹ CT-scan of the hydatid cysts has a high sensitivity of about 95-100%. It shows round and well-circumscribed cysts with low attenuation without contrast enhancement that at times may show subtle calcification.^{16,52} Mural calcification is the hallmark of HD and can be reliably detected by CT-scan.⁵⁴

Diagnosis of hydatid disease can be achieved by clinical findings, serologic tests, and imaging methods such as plain radiography, ultrasonography, CT-scan, and magnetic resonance (MR) imaging. In the majority of the reported cases, an accurate final preoperative diagnosis was confirmed by radio-imaging studies (abdominal sonography and computerized tomography) complemented with serological tests (ELISA).

Despite the aforementioned modalities, many cases with the final diagnosis of primary hydatid cysts in the above-mentioned locations were operated with the impression of lymphangioma, choledochal, pancreatic, ovarian, mesenteric, and duplication cysts.⁸ Furthermore, differential diagnoses in cases with complicated cysts were intra-abdominal abscess, hematoma, and ascites.²⁹

The gold standard for final diagnosis is histopathology in which typical cysts with scolices and hooklets are diagnostic. However, when HD is the differential diagnosis, biopsy or fine-needle aspiration are not recommended due to the risk of spillage and dissemination of the daughter cysts that can cause anaphylactic reaction and increase recurrence rate.^{45,55}

Surgical excision is still the treatment of choice. Surgery with adjuvant therapy (peri- and postoperative antiparasitic medical therapy such as albendazole) seems to remain the optimal method of treatment.²⁹ The recommended dose of albendazole is 400 mg orally twice a day for 1-5 months (pediatric dosage i: 15 mg/kg/day).

Various intra-operative scolicedal agents have also been used over the years, but the majority have shown a limited effect and produced adverse reactions. Hypertonic saline (10-15%), one of the most commonly used scolicedal agents, has been tested at various concentrations and exposure times. Therefore, careful aspiration of most of the hydatid cyst fluid before injecting the scolicedal agent into the cyst is recommended.⁴⁰

No recurrence was declared in the reported cases of peritoneal, omental, and mesenteric HD. Therefore, it seems that surgical excision with or without adjuvant medical antiparasitic disease is

a treatment of choice without complication and recurrence.⁸

Conclusion

Hydatid disease should be included in the differential diagnosis of peritoneal, mesenteric, and omental cysts in endemic countries. The most definite method for diagnosis can be perioperative ELISA test in combination with imaging studies. The treatment of choice is surgical excision in combination with peri- and postoperative antiparasitic medical therapy.

Conflict of Interest: None declared.

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