Case Report

A rare case report on the origin of hydatid cyst-how much does it matter in the management?

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ABSTRACT

Hydatid cysts are most frequently seen in the liver, followed by the lungs, spleen, and pancreas. Here, we present the case of a 17-year-old female who came with complaints of dyspnea, decreased movement on the left lower chest, and absent breath sounds. The blood test showed eosinophilia. The medical literature on hydatid cysts in the gastrohepatic space is altogether lacking. Surgical removal remains the cornerstone of treatment for large or symptomatic hydatid cysts, especially when there is a risk of rupture, compression, or secondary infection. Pre- and post-operative anti-helminthic therapy with albendazole 10 mg/kg/d ×8 weeks reduces recurrence and dissemination. This case underscores the need for a high index of suspicion for hydatid disease of gastro-hepatic space origin in patients from endemic areas presenting with respiratory complaints and its multidisciplinary approach.

Key words: Antihelminthic, Hydatid cyst, Laparoscopic surgery

Hydatid disease is a parasitic infection caused primarily by *Echinococcus granulosus*, commonly affecting populations in endemic areas who live in close contact with livestock [1]. Cystic echinococcosis is recognized as a neglected tropical disease by the World Health Organization (WHO) due to its chronic progression, disability, and socioeconomic impact [2]. The life cycle of *E. granulosus* involves domestic or wild canids, such as dogs, as the definitive hosts and herbivorous animals, such as sheep, goats, cattle, and pigs as the intermediate hosts. Adult tapeworms reside in the small intestine of definitive hosts and release eggs, which are excreted in the feces. Intermediate hosts ingest these eggs through contaminated food; these eggs hatch into oncospheres, penetrate the intestinal wall, and disseminate hematogenously to organs, most commonly the liver (about 70% of cases) and lungs (about 20–30%), where they develop into hydatid cysts [3]. Isolated extrahepatic hydatid disease most commonly occurs within the abdominal cavity, with an incidence ranging from 6% to 11% [4]. While some individuals may remain asymptomatic, the majority present with symptoms, such as abdominal pain or localized soft tissue swelling, depending on the site of involvement. Affected organs can include the spleen, pancreas, kidneys, retroperitoneum, urinary bladder, ovaries, bones, heart, thoracic wall, spinal column, thyroid gland, brain, and skeletal muscles [5]. Humans are accidental hosts, acquiring the infection through ingestion of eggs, often through contact with infected dogs. They act as dead ends in the cycle [6]. Clinical symptoms arise when cysts reach a considerable size or rupture. Hepatic cysts may cause right upper quadrant pain, hepatomegaly, or obstructive jaundice [7]. They cause mass effect, leading to elevation of the diaphragm, compression of adjacent structures, and respiratory symptoms. Pulmonary cysts may present with cough, chest pain, or hemoptysis. Cyst rupture may lead to secondary infection or life-threatening anaphylactic shock due to antigenic content release [7]. Rarely, cysts may involve other organs, such as the spleen, brain, bones, or kidneys, leading to more atypical presentations [8].

Here, we report a rare case of a giant hydatid cyst originating from the gastro-hepatic space and manifesting primarily with symptoms suggestive of thoracic involvement. This case underscores the need for a high index of suspicion for hydatid disease of gastro-hepatic space origin in patients from endemic areas presenting with respiratory complaints and its multidisciplinary approach.

CASE REPORT

A 17-year-old female, with no known comorbidities, residing in an endemic rural region, presented with mild breathlessness over 2 months. The patient was

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well oriented to time, place, and person. She denied any history of fever, cough, or weight loss. She presented with vitals of pulse rate of 108/min, blood pressure of 106/70 mmHg, respiratory rate of 22/min, with SpO_2 being 96% on room air.

On clinical examination, there was decreased movement on the left inframammary, infrascapular, and infraaxillary area with absent breath sound. On palpation of the abdomen, a firm, non-tender, and well-defined mass was palpable in the epigastric region. The mass was non-mobile with no guarding or rigidity. No hepatomegaly was noted.

Routine blood investigations were within normal limits, except for mild eosinophilia (Table 1). Clinically, pleural effusion was kept as the first possibility. A chest X-ray revealed a homogeneous opacity with a smooth, rounded, convex margin merging with the left hemidiaphragm, and a clear left costophrenic angle (Fig. 1). Ultrasound (USG) abdomen was suggestive of a cystic lesion with a thick margin above the liver in the gastrohepatic space. On further evaluation, the contrast-enhanced Computed tomography (CT) abdomen and thorax confirmed a well-defined, thick-walled cystic lesion highly suggestive of a hydatid cyst in the lung window and gastro-hepatic space (most probably the originating site, as a thickened wall is seen more in abdominal ones) (Fig. 2). The cyst caused significant displacement of the left hemidiaphragm and mediastinum, which led to dextrocardia situs solitus (levoversion).

The patient was managed with a combined approach consisting of medical and surgical management. The patient weighed around 40 kg and was given tab albendazole 600 mg/day in two divided doses for 2 weeks before surgery, consisting of pericystectomy (via laparotomy), and this was followed by tab albendazole 600 mg/day for 6 weeks. The patient improved symptomatically and was planned for review scans, which, however, was not possible due to a lack of affordability, and the patient was lost to follow-up.

DISCUSSION

Diagnosis of a hydatid cyst is based on imaging and serology. USG is the first-line diagnostic modality, especially for hepatic involvement, due to its easy accessibility and cost-effectiveness. CT and magnetic resonance imaging are valuable for evaluating pulmonary or extra-abdominal cysts. Characteristic imaging findings include well-defined cystic lesions, daughter cysts, hydatid sand, and calcifications. Serological tests, such as enzyme-linked immunosorbent assay, indirect hemagglutination, or immunoblotting, are useful adjuncts but may yield false negatives, especially in inactive or calcified cysts [9].

Hepatic hydatid cysts tend to evolve through various stages, including degeneration, calcification, and secondary infection, which can result in thickened or irregular cyst walls. Over time, the cyst wall may become fibrotic or calcified, especially in older or complicated cysts. In contrast, pulmonary hydatid cysts usually have thinner walls, as the elasticity of lung tissue allows the cyst to expand more easily without causing thickening. Thickened walls in lung cysts are less common unless there is secondary infection or complication (rupture or superinfection) [8].

Primary extrahepatic hydatid cysts are rare, and only a few sporadic cases have been reported. Here, we discuss some unusual sites of presentation of hydatid cysts mentioned in the medical literature. A 42-year-old female presented with left flank pain, and imaging revealed a hydatid cyst in the left kidney. She underwent nephrectomy. Histopathology confirmed *E. granulosus*. Post-operative albendazole therapy was initiated with no recurrence at 1-year follow-up [8]. Literature also mentions a 55-year-old male presenting with pelvic discomfort, a CT showed a multilocular cystic lesion in the pelvis. Surgical excision confirmed a hydatid cyst. Serology was positive for *Echinococcus*. The patient recovered well on albendazole [10]. There has also been a case report with mention of a hydatid cyst in the mediastinum in a 60-year-old man who complained of chest pain. Chest CT revealed a cystic mass in the mediastinum. Surgical removal confirmed hydatid disease. Eosinophilia and serology supported the diagnosis. Albendazole was prescribed post-operatively [11].

Medical literature on hydatid cysts in the gastro-hepatic space is altogether lacking. Although hepatic hydatid cysts are relatively common in endemic areas, giant cysts in the left lobe are rare and can pose

Table 1: Laboratory parameters of the patient

Lab parameter	Patient value	Reference range
Hemoglobin (g/dL)	12	12–17
Total leucocyte count (cells/mm^3)	12000	4000–11000
Differential leukocyte count granulocytes/lymphocytes/eosinophils (%)	66.2/23.8/10	40–80/20–40/1–6
Platelet count (lac cells/mm^3)	2.35	1.5–4
Erythrocyte sedimentation rate (mm/1 h)	14	<10
S. Sodium/potassium/ionized calcium (mmol/L)	138.1/4.5/1.20	135–150/3.5–5.3/1.13–1.32
Serum glutamic-oxaloacetic transaminase/serum glutamic pyruvic transaminase/alkaline phosphatase (U/L)	44/49/284	<40/<45/<270
S. protein/albumin/globulin (g/dL)	7.1/3.5/3.6	6.4–8.3/3.5–5.2/2–3.5
S. urea/creatinine (mg/dL)	25/0.89	13–43/0.8–1.3
S. Bilirubin (total/direct/indirect) (mg/dL)	0.36/0.16/0.2	<1.2/<0.2

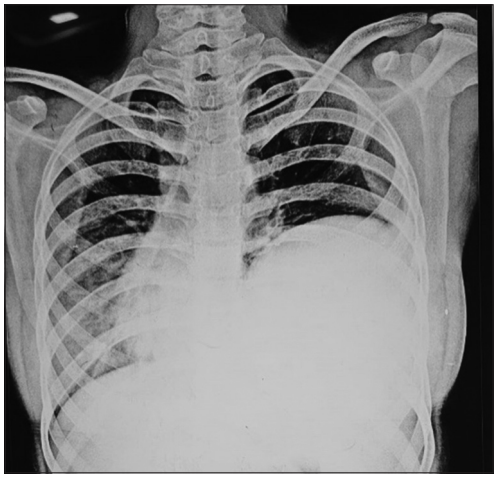

Figure 1: Chest X-ray posteroanterior view revealing homogenous opacity with clear left costophrenic angle

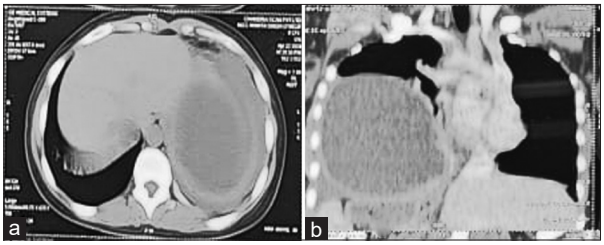

Figure 2: (a) Contrast-enhanced computed tomography (CECT) abdomen showing thick-walled cyst in the gastrohepatic space; (b) CECT thorax confirming thick-walled cyst in the infradiaphragmatic area pushing the heart to the right

diagnostic and therapeutic challenges. Their clinical presentation may be misleading, as in this case, where the primary concern was breathlessness due to altered hemidiaphragmatic contractility, compromised lung, and dextrocardia. Differential diagnoses included were left-sided pleural effusion (may be loculated as the margin was convex upward), hydatid cyst, subdiaphragmatic abscess, splenic cysts, or even thoracic masses. The patient had no history of pancreatitis, which is mostly a pre-cursor to pancreatic pseudocyst. USG did not reveal any gallstones either. Thus, the possibility of a pancreatic pseudocyst was ruled out.

Treatment options depend on cyst location, size, stage, and patient condition. They include surgery (particularly for complicated or large cysts), percutaneous techniques, such as puncture, aspiration, injection, and reaspiration (PAIR), and antihelminthic therapy with albendazole or mebendazole. The WHO Informal Working Group on Echinococcosis has classified cysts into active, transitional, and inactive stages to guide therapy. Surgical removal remains the cornerstone of treatment for large or symptomatic hydatid cysts, especially when there is a risk of rupture, compression, or secondary infection. Pre- and post-operative anti-helminthic therapy with albendazole 10–15 mg/kg body weight/day for 8 weeks reduces recurrence and the risk of dissemination. The PAIR technique or laparoscopic approaches may be considered depending on cyst location and expertise availability [12]. Prevention involves a "One Health approach," focusing on deworming of dogs, safe disposal of infected feces, meat inspection, and community education in endemic areas [2].

CONCLUSION

Hydatid cyst disease remains a major public health concern in endemic regions. Primary extrahepatic hydatid cysts are rare, and only a few sporadic cases have been reported. Gastrohepatic space is not directly involved in this filtration route; hence, primary involvement is rare. This case underscores the need for a high index of suspicion for hydatid disease of gastro-hepatic origin in patients from endemic areas presenting with respiratory complaints. Unusual presentations, such as elevation of the hemidiaphragm due to massive cysts, should prompt detailed imaging of the abdomen as well to avoid misdiagnosis. Early recognition and appropriate therapy can lead to favorable outcomes and prevent serious complications, such as rupture or anaphylaxis. Multidisciplinary strategies combining early diagnosis, appropriate treatment, and preventive veterinary and human health measures are essential to reduce its global burden.

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