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*Case Report*

# Disseminated lung hydatidosis from intraabdominal hydatid cyst via inferior vena cava (IVC): a very rare presentation

Mahmoud Yousefi-Mashhour[1], Shora Shafighnia[2], Zahra Taherzadeh[2], Farahnaz Joukar[2], Fariborz Mansour-Ghanaei[1]

*[1]Division of Gastroenterology & Hepatology, Gastrointestinal & Liver Diseases Research Center (GLDRC), Guilan University (Medical Sciences), Rasht, Iran; [2]Gastrointestinal & Liver Diseases Research Center (GLDRC), Guilan University (Medical Sciences), Rasht, Iran*

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**Abstract**: Hydatid disease is a parasitic infestation by Echinococcus granulosus characterized by cyst formation in various organs. Secondary involvement due to hematogenous dissemination may be found in almost any anatomic location. We herein reported a very rare case of lung hydatidosis as a result of disseminated hydatid cysts from intra-abdominal through IVC in a young female patient. She was treated with surgical intervention in two stages followed by Albendazole therapy.

**Keywords:** Lung hydatidosis, intraabdominal, hydatid cyst, inferior vena cava

## Introduction

Hydatid disease is a serious medical problem in Mediterranean and particularly among sheep-farming countries, caused by larval stages of dog tapeworms belonging to the genus Echinococcus. Hydatid cyst may affect every organ in human body, however peritoneal echinococcosis is uncommon. It may be asymptomatic or may lead to lethal complication [1, 2]. Diagnosis of hydatidosis is based on immunodiagnostic methods along with radiological and ultrasound examinations [3].

## Case report

A 23 year old female, carpet weaver from a rural area, in north of Iran (Guilan province), referred to our gastroenterology department at Razi hospital, complaining of new onset acute abdominal pain, 2 years duration of cough with blood tinged sputum.

She suffered from a non-productive cough, occasional fever of two years' duration and also from abdominal pain of 5 months ago. Her abdominal pain was mild and constant, unrelated to meals, aggravated after cough, radiating to back. At no time was there any symptoms such as nausea or vomiting, jaundice, change in urine or stool color. She had significant weight loss (8.2 kg in 1 month). Her past medical history revealed only Iron deficiency anemia. Her social history about smoking and drinking alcohol was negative and her family history was also not significant aspects. She had history of exposure to dog and sheep occasionally.

On physical examination, she was pale and weak. Her vital signs were stable and afebrile. (BP: 100/70, PR:78, RR:18, T:36.9). At abdominal examination, her abdomen was soft, tender to palpation of the RUQ, with no distention or organomegaly and no rebound tenderness or guarding. The other results of physical examination were normal.

In lab test, her hemoglobin was 10 ml/dl. Her leukocyte count was 8900/umm that consist of 40% polymorphnuclear, 38% lymphocyte, 20% eosinophil and 2% monocyte. Rest of the routine laboratory investigations, including liver and

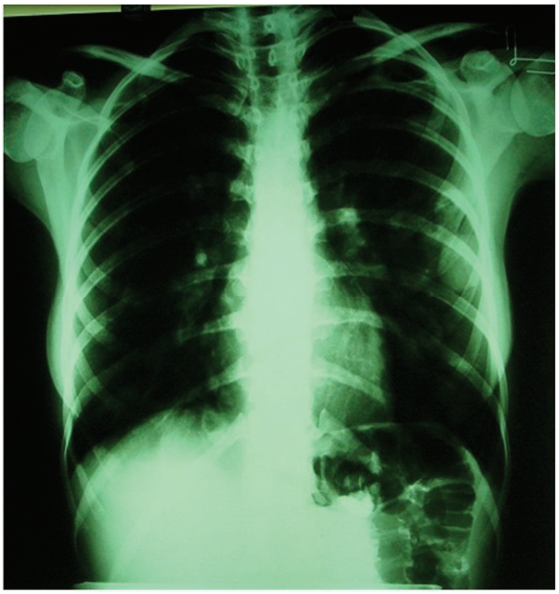

**Figure 1.** The posterior anterior chest radiograph showing bilateral multiple pulmonary nodules.

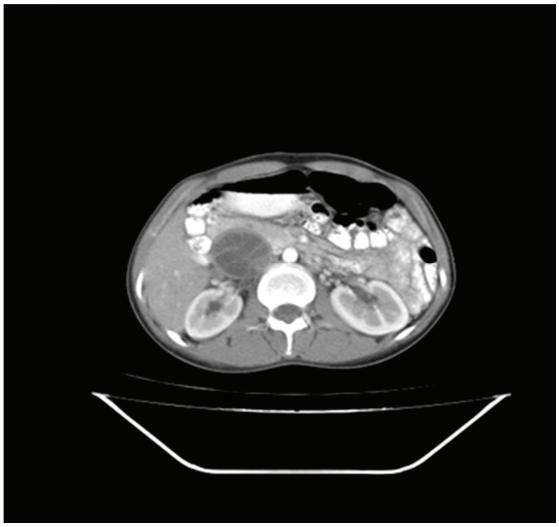

**Figure 2.** First Abdominal CT scan. Retroperitoneal multicystic mass in posterior aspect of pancreatic head.

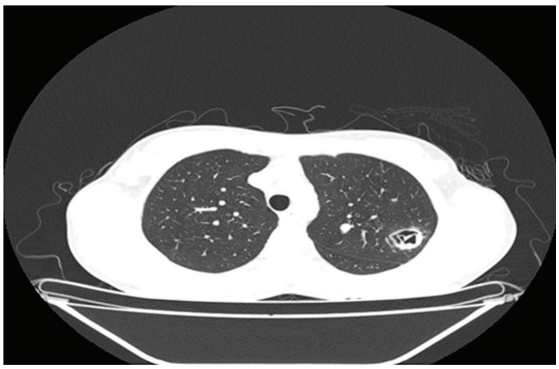

**Figure 3.** Chest CT-Scan revealed a cyst in the left middle lobe of the lung containing a freely floating endocyst (the "water lily sign").

kidney function tests, serum proteins, urinanalysis and stool exam were normal. The erythrocyte sedimentation rate (ESR) was 67 mm/hr (Normal<20 mm/hr). Moreover, the patient had negative reaction of the Mantoux tuberculin skin test. Her Chest X-ray showed multiple nodules and cystic lesion in both lobes (**Figure 1**).

CT (computed tomography) of her thorax and abdomen revealed retroperitoneal multicystic mass measured approximately 105 X 52 X 37 mm in posterior aspect of pancreatic head that extends adjacent to right kidney and posterior of right portal vein and hepatic artery and result in compression and collapse of inferior vena cava (IVC) (**Figure 2**). In addition, a cyst was revealed in the left middle lobe of lung containing a freely floating endocyst (The water lily sign, is pathognomonic for Echinococcus) (**Figure 3**). Serological testing to Echinococcus by enzyme-linked immune-sorbent assay (ELISA) confirmed the diagnosis with high titers of IgG (108.5 > 15 is taken as positive). Oral albendazole was started as soon as at the dose of 400mg twice daily and she then referred to surgical ward for thoracotomy and laparotomy. Unfortunately, our patient rejected thoracotomy, she was unwilling to simultaneously thoracic and abdominal surgery. At laparotomy abdominal mass was excised. During the operation, it was observed that the lesion had also involved the IVC. Gross pathological examination revealed a cystic soft mass containing many daughter cysts (**Figure 4A, B**). Also, histopathology demonstrated the presence of a soft cystic mass filled with gelatinous material and multiple daughter cysts. Following treatment with surgery accompanied by albendazole, the patient was discharged in a healthy condition on the ninth postoperative day with a prescription of albendazole 400mg twice a day for a period of 6 months. Respiratory symptoms recurred 3 months after discontinuation of albendazole. During this period, abdomi-

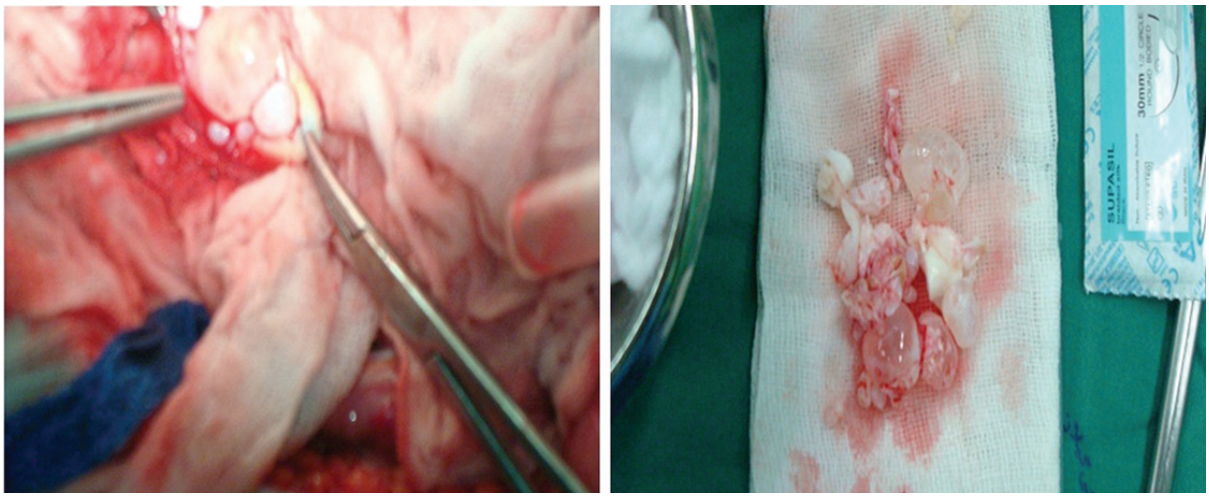

**Figure 4.** A: Surgical appearance of the mass exposed; B: Gross pathological specimen of hydatid cyst with multiple daughter cysts removed from intra-abdominal space.

nal symptoms have entirely subsided. A follow-up thoracoabdominal CT-scan with intravenous contrast was performed 6 months later. CT-scan of her abdomen was normal and there is no evidence for mass or lymphadenopathy. But CT scan of her chest showed multiple nodules with various size (**Figure 5**). A planned thoracotomy was chosen for this patient. Posterolateral thoracotomy through the sixth intercostals space was accomplished with the patient in the lateral decubitus position. Cystotomy, capitonnage plus decortication of the pleura, was carried out in our patient. In addition, the resection of the sixth costal was done for osteomyelitis. We prescribed albendazole regimen in our case regarding multiple hydatid cyst due to probable recurrence in the postoperative period. Her respiratory symptoms subsided following the surgical procedure. The next follow-up examination is recommended 3 months later.

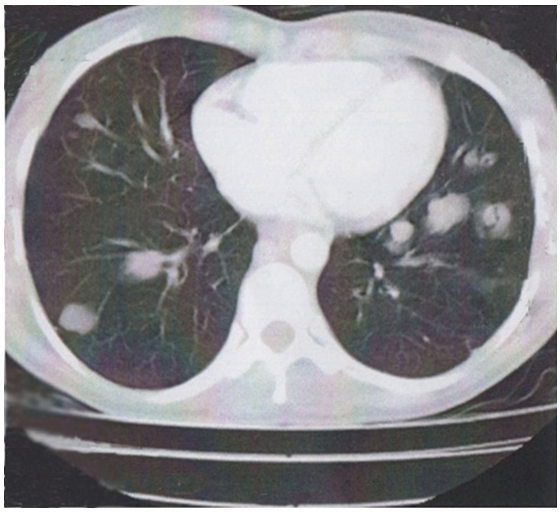

**Figure 5.** The second Chest CT-Scan 6 months later. Multiple cyst hydatidosis in her lungs.

## Discussion

Echinococcus granulosus is a cosmopolitan parasite. The overall incidence of Echinococcus infection is 0.4 per 100,000 persons [1]. It is a unique parasitic disease which can occur almost anywhere in the body and demonstrates a spectrum of imaging features that vary according to growth stage, associated complications, and affected tissue [4]. Hydatid cyst can be found rarely in the peritoneum [5]. Moreover, Hydatid cyst may distribute into more space and produce a lot of daughter cysts like our case. The exact mechanism how the larvae pass through the liver and lungs and form solitary cyst elsewhere is not well understood. It is suggested that lymphatics are responsible for systemic dissemination of the larvae and formation of solitary cysts at uncommon sites Patients with hydatid disease are mostly asymptomatic [6], but perforation into the abdominal cavity may result in hazardous complications causing anaphylaxis and sudden death [7]. Multiple or-

gans involvement is also seen many studies [8], this state as seen in our case, lung and intra-abdominal cavity both were involved by hydatid cyst. Involvement of lung by hydatid cysts in this patient was due to compression of the IVC and disseminated daughter cysts from intra-abdominal via IVC. The diagnosis is based on the history of exposure in an endemic area, ultrasonography and Computed Tomography findings. The diagnosis can be supplemented by specific IgG, complement fixation, indirect fluorescent, and ELISA tests. The severity of various serological tests used for hydatid disease varies from 64 to 87% [9]. However, rarely hepatic or intra abdominal cysts may rupture into hepatic veins or inferior vena cava with subsequent embolization of the cyst contents into the lungs as it appears in this case [10].

Overall the management of both primary and recurrent hydatid disease is surgical, as anti-helminthic chemotherapy alone has failed in many cases. However, The World Health Organization recommendations state that medical therapy should be used for: patients with inoperable disease, patients with multiple cysts in two or more organs, patients with peritoneal cysts, patients following incomplete surgery or relapse and for prevention of secondary spread of echinococcal infection following spontaneous rupture or aspiration of cysts [11]. We believe the recurrence in our case is due to wrong decision of patient as to reject thoracotomy and following to stop medical therapy. Nevertheless albendazole is the most commonly used drug for pre- and post-operative antihelminthic treatment and continued suppressive therapy of inoperable hydatid cyst [12]. This report corroborates the ideas of Ghoshal et al, who found surgery was the most effective way of treatment of thoracic hydatid cyst along with preoperative albendazole therapy [13].

## Conclusion

We concluded that echinococcal disease should be taken into consideration in the differential diagnosis of every cystic mass in any anatomic location. Furthermore, Conservative surgical procedures should be used as first choice and an appropriate surgical approach results in low complication and recurrence rates.

**Address correspondence to:** Dr. Fariborz Mansour-Ghanaei, Gastrointestinal and Liver Diseases Research Center (GLDRC) Razi hospital, Sardarjangal Ave. Rasht, Iran. Tel: +98-131–5535116; Fax: +98-131-553495; E-mail: ghanaei@gums.ac.i or ghanaie@yahoo.com

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