



# Hydatidosis of the liver and posterior mediastinum

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## ABSTRACT

**INTRODUCTION:** Cystic echinococcus (CE) is an endemic zoonosis secondary to infection by the larval form of the cestode *Echinococcus granulosus*. An intermediate host, humans enter the organism's life cycle by exposure to infected canid feces. The liver is the most common location of CE while mediastinal hydatid cysts are rarely reported.

**PRESENTATION OF CASE:** We report a case of synchronous CE of the liver and posterior mediastinum treated sequentially using chemotherapy, percutaneous aspiration with injection of a scolicalid agent and re-aspiration (PAIR) and then staged minimally-invasive surgeries.

**DISCUSSION:** Synchronous CE involving the liver and posterior mediastinum is rare. The treatment of hydatid liver and mediastinal disease is multimodal including chemotherapy, percutaneous and laparoscopic or open surgical interventions. One option for controlled puncture of hepatic and mediastinal CE includes PAIR followed by surgery.

**CONCLUSION:** The sequential use of chemotherapy and PAIR followed by surgery provides another treatment strategy for management of CE. We believe this strategy may be used safely in locations without endemic CE, including most regions of the United States.

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## 1. Introduction

CE is a parasitic disease endemic to many parts of the world involved in sheep-raising causing a significant public health problem. The disease may affect many different organs, involving the liver most commonly followed by the lung. Treatment of CE may include chemotherapy, percutaneous aspiration, injection of scolicalid agent and re-aspiration (PAIR) or surgery [1].

We present a case of synchronous CE of the liver and posterior mediastinum treated sequentially with chemotherapy and PAIR to both lesions followed by partial cystectomies via minimally-invasive techniques.

## 2. Presentation of case

A 44-year-old man presented with right upper quadrant abdominal pain. He was without significant past medical or surgical history. He denied any farm exposures, never worked with livestock but had dogs as pets throughout his life. He was born in Los Angeles, California, and entered the Navy; subsequently traveling overseas to Bahrain, Dubai and Mexico, last visiting in 2007.

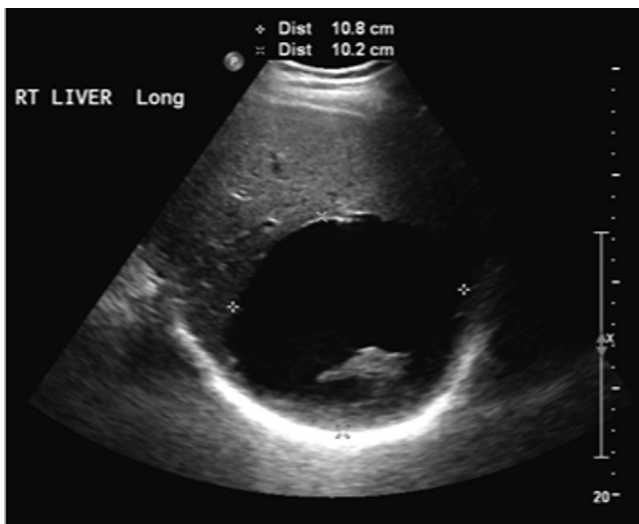
His physical examination was normal as were his liver function tests except for elevated alkaline phosphatase of 131 U/L. A right

upper quadrant ultrasound demonstrated a hypoechoic 10 × 10 cm focus within the right liver lobe with a heterogeneous, solid interior focus without flow. This focus was described as well-circumscribed and thin-walled; the cyst wall was without vascularity (Fig. 1). A liver MRI was obtained which demonstrated cystic lesions not only within the right lobe of the liver and but also in the paraspinal region in the left lower thorax. The thoracic cyst was noted to be 11 × 6 cm and had similar signal characteristics to the liver cyst (Fig. 2). No CT scan was obtained.

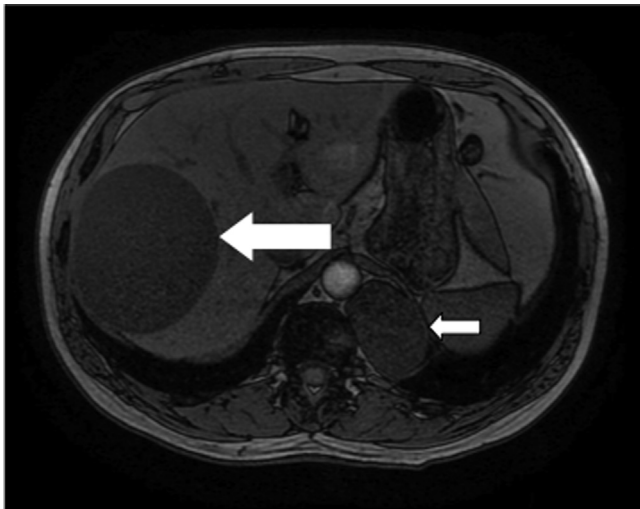
Given the radiologic characteristics, the most likely diagnosis was CE even though *Echinococcus* serologies were negative. Cyst excision was planned for definitive histopathologic diagnosis and treatment. He was started on a seven-day course of albendazole and then he underwent PAIR to both his hepatic and mediastinal cysts with 20% hypertonic saline by Interventional Radiology. After injection of contrast within the hepatic cyst, no biliary transit was noted. The smears from the aspirations were suggestive of hydatid sand; however no hooklets or protoscolices were identified.

One day after his PAIR procedure, he underwent a laparoscopic hepatic partial cystectomy and omentoplasty. Prior to incision, 20% hypertonic saline was injected into the hepatic cyst through the percutaneous drain. Intra-operatively, sponges infiltrated with 20% hypertonic saline were placed surrounding the liver cyst. A large right hepatic cyst along the dome of the liver was then unroofed using a Harmonic scalpel (Fig. 3). A tongue of omentum was then placed within the cavity and sutured into place. Cystic wall tissue sent for histopathology was compatible with *Echinococcus* via direct

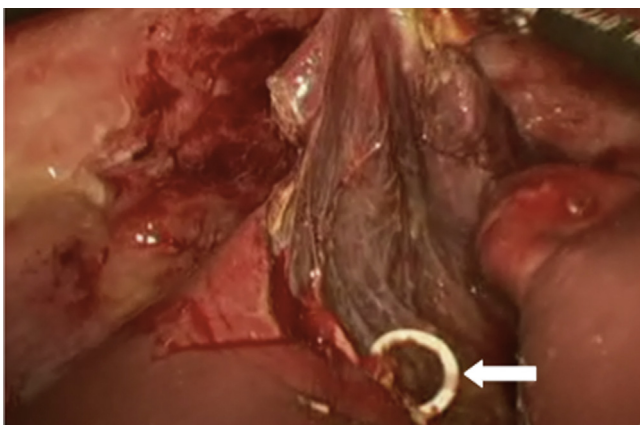
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**Fig. 1.** Right upper quadrant ultrasound demonstrated a 10 × 10 cm rounded, thin-walled cyst containing a heterogeneous focus without flow.



**Fig. 2.** MRI demonstrated not only the liver cyst (large arrow) but also a 10 × 6 cm posterior mediastinal cyst (small arrow) without any communication to the neural foramina.



**Fig. 3.** Hydatid cyst being unroofed laparoscopically, note pigtail catheter (arrow) in the cyst cavity placed the day before by Interventional Radiology.

visualization of protoscoleces and hooklets. Two days following his first operation he underwent a left video-assisted thoracoscopy (VATS) and pericystectomy of his posterior mediastinal cyst. After dividing the inferior pulmonary ligament and placing 20% hypertonic saline-soaked sponges in the posterior mediastinum, the cyst was dissected off the descending thoracic aorta, posterior chest wall and diaphragm. The cyst did not involve the lung parenchyma. The mediastinal cyst tissue was also compatible with *Echinococcus*, demonstrating an outer cyst wall composed of acellular hyalinized fibrous tissue with focal calcifications. The patient tolerated the procedure well and was discharged to home a few days following his thoracic procedure.

A 6-month course of albendazole was then completed. Infectious disease follow-up surveillance subsequently included repeat abdominal/pelvis CT scans every 3–6 months for a minimum of 3–5 years given the dual location of the hydatid cysts.

### 3. Discussion

CE caused by *Echinococcus granulosus* has a worldwide distribution with an incidence of 1–200 cases per 100,000 people, the highest being found in endemic regions such South America, Russia, China, Eastern Europe and the Mediterranean countries [1,2]. Given that this disease is not officially reportable in the United States, epidemiologic data here has been scant. CE does occur in North America and has been known to be endemic to California [3]. More recently, however, Moro et al. reported that autochthonous transmission in California appeared to have ceased given that most cases were in immigrants from endemic countries [4]. However, given his lack of travel to areas endemic to hydatid disease, our patient may have become infected via autochthonous transmission in California.

The most common location of CE is the liver, affecting approximately 75% of cases while finding CE in the posterior mediastinum is rarely reported [5]. As of 2002, Eroglu et al. reported just 100 case of mediastinal CE in the English literature [5]. Among intrathoracic CE, the incidence of mediastinal CE is about 0.1–0.5% [6]. When discovered in the mediastinum, CE is most common in the posterior mediastinum. In their series of 74 patients, Rakower and Milwidsky reported a 55% rate of primary cysts in the posterior mediastinum, 36% in the anterior mediastinum and <8% in the middle mediastinum [6]. Synchronous pulmonary and hepatic CE may occur in 4% to 25% of cases [7], whereas synchronous posterior mediastinal and liver CE is rare. Of 7 cases involving mediastinal hydatidosis, Kabiri et al. reported one case of synchronous mediastinal and liver CE; otherwise the literature is scarce describing such cases [8].

Given the proximity of vital structures in the mediastinum, surgical management is recommended [5]. For intrathoracic extrapulmonary hydatid cysts, total removal of the germinative membrane and pericyst has been encouraged [9]. However, the proximity and adherence to vital mediastinal structures may necessitate partial resection precluding pericystectomy. In these cases where preoperative evaluation suggests that pericystectomy may not be performed safely, preoperative PAIR may allow for controlled puncture and prevention of pleural seeding and/or anaphylactic reactions.

As with his posterior mediastinal cyst, we treated his hepatic CE with albendazole followed by sequential PAIR and laparoscopic partial cystectomy. The treatment of hepatic CE may involve the use of PAIR or surgery (open or laparoscopic) with or without chemotherapy [10]. One of the initial concerns using laparoscopy to treat hepatic CE was puncturing the cyst under high-pressure, potentially leading to peritoneal seeding and/or anaphylaxis. In a review of 8 case series involving 334 patients where laparoscopy was used to manage hepatic CE, 3 cases of anaphylaxis were reported [10]. These procedures were completed by laparoscopic

surgeons experienced with hydatid surgery therefore the risk of anaphylaxis may be greater with those less experienced. For these reasons, numerous laparoscopic techniques have been developed for controlled puncture and aspiration [11–13]. In areas not endemic to CE, where it would not be cost-effective to obtain such suction-grinders or special trocars, PAIR prior to surgical management may be pursued.

#### 4. Conclusion

Hepatic and posterior mediastinal CE presenting simultaneously is rare. To allow for controlled puncture of cysts in both locations prior to operative management, PAIR was completed. We believe treating CE with chemotherapy and PAIR followed by surgical intervention provides another effective approach to the treatment of CE. This treatment paradigm allows for controlled-puncture prior to surgical intervention with prevention of possible seeding and/or anaphylaxis.

#### Conflict of interest

No conflicts of interest exist.

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#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Author contribution

JQ: Study design, data collection and analysis, writing.  
 DG: writing.  
 WD: Study design, writing.

#### Disclaimer

The views expressed herein are my own and do not necessarily reflect the official policy or position of the Department of the Navy, Department of Defense, or the U.S. Government.

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