



## Case report

## Secondary disseminated intraperitoneal hydatid cyst: A case report

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## ARTICLE INFO

## Keywords:

Hydatidosis  
Echinococcosis  
Cystic echinococcus  
Hydatid cyst  
Case report

## ABSTRACT

**Introduction:** Hydatidosis is an uncommon zoonotic infection in the Philippines with only a few reported cases. It frequently presents as benign liver cysts with variable symptoms often related to mass effect.

**Case presentation:** We present a 49-year-old male with a significant travel history from East and Central Asia, surgically treated twice as a benign liver cyst and now presenting with recurrent multiple intraabdominal cysts. He subsequently underwent surgical excision of multiple hydatid cysts with prolonged albendazole treatment. The patient presently remains disease-free as of most recent follow-up.

**Discussion:** The disease is caused by the tapeworm *Echinococcus granulosus* and transmission happens when humans acting as intermediate host ingest food contaminated with the parasite eggs, resulting to the liver being the most common location. Frequently, it manifests and is treated as incidental hepatic cysts for the disease is characterized to have a long asymptomatic period. Symptoms are variable and may range from having abdominal pain, increasing abdominal girth, vomiting, fever or myalgia. Ultrasound or CT scan reveals single or multiple cysts however, biopsy of the cyst wall with demonstration of the larval form (protoscoleces) is diagnostic.

**Conclusion:** This case highlights the value of history-taking and having a high-index of suspicion particularly for rare cases.

## 1. Introduction

Hydatid disease (also known as hydatidosis, cystic echinococcus or echinococcosis) is a zoonosis that has a low prevalence in Southeast Asia, particularly in the Philippines, the epidemiology of which is characterized as extremely uncommon. There have been only 5 reports of echinococcosis (2 in humans, cattle, water buffalo and dog) since 1925, all of which were located in the lungs [1]. The disease has a long asymptomatic period (>5 years) which makes it difficult to estimate the actual numbers in endemic regions. The World Health Organization (WHO) reports that more than a million people are affected at any one time with the infection being prevalent in areas where there is close contact with dogs and livestock [2]. Symptoms are variable and commonly present once the cysts have grown so large as to cause mass effect or have involved adjacent organs. Ultrasound (US) frequently reveals cystic lesions in the liver or lungs and treatment is often

expensive, at times complicated requiring extensive surgery or prolonged drug therapy. Prevention programs in place include deworming dogs, slaughterhouse hygiene and public education. Herein we describe the first reported case of disseminated peritoneal hydatidosis in the country which happened as a result of a surgical complication. This case presents an opportunity to emphasize the diagnostic dilemma, importance of history-taking and discuss options in management of such a rare case. This report has been written in line with the recent SCARE criteria for case reports [3].

## 2. Case

A 49-year-old male presented with a 3-month history of gradual abdominal enlargement, flank pain, fever and diarrhea. Condition started 4 years prior when he experienced easy fatigability and myalgia travelling back from Nepal. During work-up for a mitral valve

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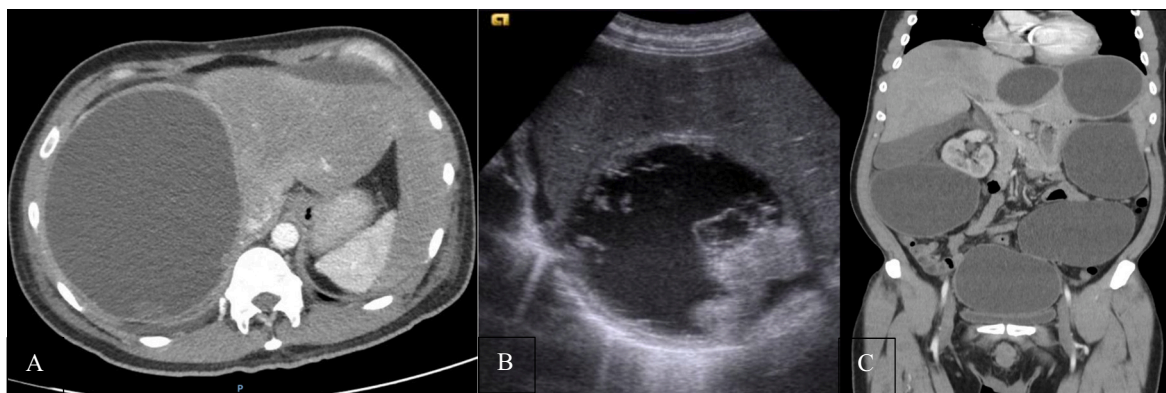
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<https://doi.org/10.1016/j.ijscr.2022.107036>

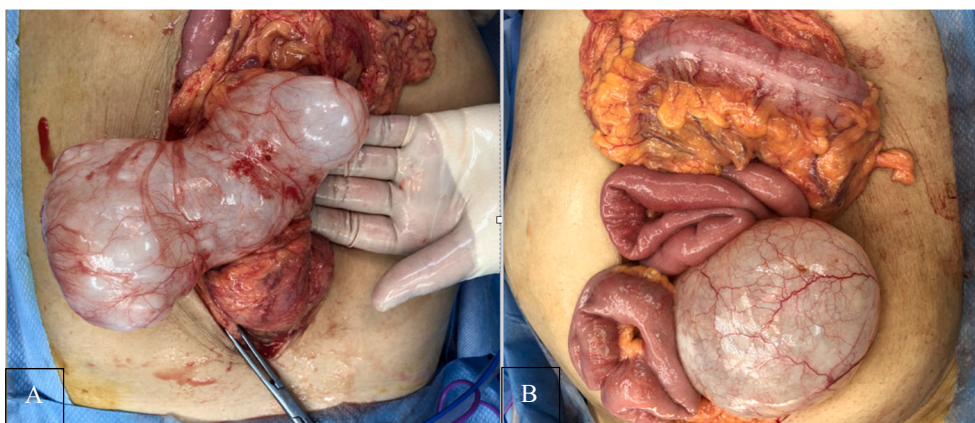
Received 10 March 2022; Received in revised form 2 April 2022; Accepted 2 April 2022

Available online 7 April 2022

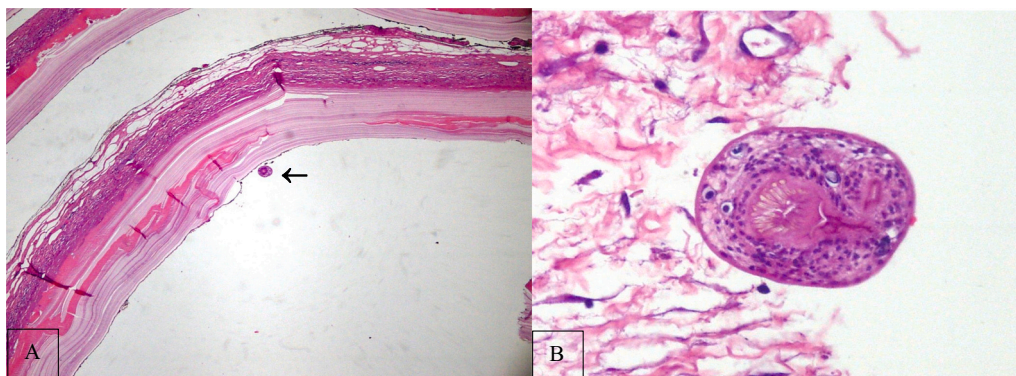
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**Fig. 1.** Initial CT scan showing huge right hepatic lobe cyst (A). Ultrasonographic findings of complex hepatic cysts 2 years after initial surgery (B). CT scan findings of recurrent, multiple abdominopelvic cysts after 4 years and 2 laparoscopic surgeries (C).



**Fig. 2.** Intraoperative findings showing varisized cysts. Shown here is a infrahepatic cyst measuring 16 × 20 cm (A) and a sigmoid cyst measuring 15 × 15 cm (B).



**Fig. 3.** Hematoxylin and Eosin (H & E) stain, original magnification ×20 – Low-power microscopic view of the hydatid cyst wall showing three layers (A). The outermost adventitial layer is composed of fibrovascular tissue with focal, mild lymphocytic infiltrates, followed by an acellular, hyaline, laminated membrane. Innermost wall (germinal layer) is lined by a thin, nucleated layer. On the luminal side, one protoscolex is noted (arrow). H & E stain, original magnification ×200 – High-power microscopic view demonstrating parts of protoscolex with refractile hooklets and a round sucker (B).

replacement, he was noted to have an incidental right hepatic lobe cyst measuring 15 × 12.5 × 17 cm on computed tomography (CT) scan for which a laparoscopic marsupialization was performed a few months after his cardiac surgery (Fig. 1A). Biopsy result showed bile duct cyst and eosinophilic amorphous materials. The patient was asymptomatic until after 2 years, when he presented with epigastric pain and recurrence of the hepatic cysts. Ultrasound at that time revealed multiple complex bilateral hepatic lobe cysts and gallstones (Fig. 1B). He again underwent laparoscopic marsupialization with cholecystectomy. Biopsy was consistent with simple hepatic cyst. He remained asymptomatic only for the symptoms to recur after 2 years. Latest CT scan showed

multiple, well-defined, varisized, hypoattenuating abdominopelvic cysts causing mass effect on the visceral organs (Fig. 1C). He subsequently underwent exploratory laparotomy and excision of the numerous cysts (Fig. 2A, B). Final histopathology revealed hydatid cyst for which albendazole (10 mg/kg) 400 mg BID for 6 months was initiated (Fig. 3A, B). His postoperative course was unremarkable and presently with no recurrence after six months of follow-up.

### 3. Discussion

Hydatidosis is a zoonotic infection caused by *Echinococcus granulosus*

which is endemic in many regions such as East Africa, Central Asia and China with a prevalence rate of 5–10% [2]. Humans are considered accidental intermediate hosts and are infected through ingestion of tapeworm eggs in contaminated food, water or soil or direct contact with the definitive animal hosts (i.e., dogs). Hydatid cysts developed frequently in the liver (80–90%), most commonly the right lobe (50–70%) as the larvae (metacestodes) released penetrates the intestines and spreads hematogenously [4]. Because of its slow growth, patients may be asymptomatic for years until these cysts enlarge and cause compressive symptoms. The innermost germinal layer of the cyst generates brood capsules and protoscoleces into a central cavity filled with a clear “hydatid fluid” [5]. Infrequently, cysts may spontaneously rupture and induce anaphylaxis or lead to secondary disseminated cystic echinococcosis. Disseminated echinococcosis can also arise as a complication of surgery for hydatid disease as presented in this case. Despite the extensive travel history from an endemic area, the patient was surgically treated twice as a benign liver cyst. Marsupialization or unroofing of the cysts most likely led to the spillage and seeding of its contents intra-abdominally causing peritoneal hydatidosis. Peritoneal hydatidosis is an uncommon disease with few cases reported in the literature. Kosmidis et al., reported the largest series of 34 peritoneal hydatidosis who underwent various surgeries ranging from pericystectomy, aspiration or excision with or without application of scolicalidal agents albeit with a recurrence rate of 23% [6]. Unusual presentations include intestinal ischemia, enteric fistulas and fever of unknown origin [7].

Ultrasound or CT scan remains to be the diagnostic modality of choice with lesions appearing well-defined and at times multiseptated. CT scan has a higher reliability particularly for determining relationship to nearby structures and assessing other extrahepatic cysts while MRI can better delineate liquid areas within the cyst matrix. In addition, other modalities like antibody assays such as enzyme immunoassay, latex agglutination and indirect hemagglutination may be utilized although with lower sensitivities [8]. Demonstration of protoscoleces on microscopic examination after cyst fluid aspiration may also be diagnostic.

There are various acceptable treatment approaches which are based on the recent WHO classification of hydatid disease mainly medical treatment with anti-helminthics (albendazole, praziquantel, or ivermectin), percutaneous aspiration, excision or even watchful waiting [9]. However, it is less clear for disseminated peritoneal hydatidosis. Although surgical excision of all visible cysts is most preferred, some have advocated inactivation of liver protoscoleces within the cyst before excision by washing with 20% hypertonic saline as a scolicalidal agent. Neoadjuvant anti-helminthic therapy has also been recommended consisting of 2–3 weeks of albendazole treatment prior to the contemplated procedure followed by 6–8 weeks more after surgery [10]. Unfortunately, in this case, the surgeon was not highly considering hydatidosis when he performed the initial 2 surgeries, hence no neoadjuvant therapy nor scolicalidal agent were given intraoperatively. The surgical approach, however, will depend on the number, size and location of the cysts, nature of complications and previous surgery. Laparoscopy may be an option in selected cases, but the risk of seeding or spillage has never been fully evaluated. Overall mortality has been reported to be relatively low (2–4%) and is usually due to the development of complications [11].

#### 4. Conclusion

In conclusion, our case describes a patient who underwent minimally invasive surgery for a presumed benign and simple liver cyst wherein there was a low index of suspicion for echinococcosis. As a complication, there was spillage of parasitic elements leading to disseminated hydatidosis for which he successfully underwent surgery with no recurrence at most recent follow-up. Adjuvant medical treatment given was albendazole for 6 months. This case further highlights the significance and importance of careful history taking in every patient encounter.

#### Provenance and peer review

Not commissioned, externally peer-reviewed.

#### Sources of funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

#### Ethical approval

Yes.

#### Consent

Yes.

#### Author contribution

Derick Cabahug. MD: writing of paper, draft preparation.  
Earl Nathaniel Cruz MD: data collection, draft preparation.  
Bernice Navarro MD: study concept, writing of paper.  
Narciso Navarro, MD: draft preparation, data collection.  
Siegfredo Paloyo, MD, MPH: final editing, supervision, reviewing.

#### Research registration

N/A.

#### Guarantor

Siegfredo R. Paloyo, MD, MPH.

#### Declaration of competing interest

None.

#### Acknowledgments

None.

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