



Tehran University of Medical
Sciences Publication
<http://tums.ac.ir>

Iran J Parasitol

Open access Journal at
<http://ijpa.tums.ac.ir>



Iranian Society of Parasitology
<http://isp.tums.ac.ir>

Case Report

An Interesting Case of Hydatid Cyst of Liver Concomitant with Hydatid Cyst of Broad Ligament: A Case Report

Tharun Chitrambalam, *Nidhi George, Ramyasree Paladugu, Sundeep Selvamuthukumar, Amit Gilani

Department of General Surgery, SRM Medical College Hospital and Research Centre, Chennai, India

Received 17 Aug 2022

Accepted 19 Nov 2022

Keywords:

Hydatid cyst;
Echinococcus;
Parasite;
Unusual;
Location

*Correspondence Email:
drnidhigeorge@gmail.com

Abstract

Hydatid cyst is an anthrozoonotic disease caused by *Echinococcus* for which man is an accidental intermediate host. The hydatid disease commonly involves the liver and lungs. Involvement of extrahepaticopulmonary sites is extremely rare and only a few isolated cases have been reported. In 2022, a 49 year old female from the southern part of Indian subcontinent presented to us with recurrent hydatid cyst of liver co-existing with hydatid cyst of the left broad ligament, twenty years following the initial procedure. She underwent exploratory laparotomy and cystectomy and was then managed by ERCP and stenting following which she is asymptomatic till date. Though there are no hard and fast rules, the management of such cases mandate proper exploration to avoid any recurrence. Tailored surgical approaches maybe required according to the patient condition for effective, safe and recurrence free treatment of hepatic hydatidosis.

Introduction

Hydatid cyst has been described by Hippocrates as “liver full of water.” (1). This is a zoonotic disease caused by *Echinococcus granulosus*, *E. multilocularis* and *E. oligarius* endemic in sheep grazing countries like India, Australia, Middle East, Africa and South America and is transmitted by ingestion of eggs (2, 3). The frequent target

organ of this parasite is the liver via the portal tract followed by the lung. Rare localisations have been reported in the spleen, skeleton, kidney, brain, cardiac muscle and subcutis (4). Pelvic organs in females are rarely the site of cyst formation. Bickers et al reviewed 532 case of hydatid disease from an endemic area over a period of twenty years and only two cases of



Copyright © 2023 Chitrambalam et al. Published by Tehran University of Medical Sciences.
This work is licensed under a Creative Commons Attribution-NonCommercial 4.0 International license.
(<https://creativecommons.org/licenses/by-nc/4.0/>). Non-commercial uses of the work are permitted, provided the original work is properly cited

these were reported to have cysts in the broad ligament (5). It accounts for 0.3% of all gynaecological laparotomies in an endemic area (6).

Surgery is the treatment method of choice and includes multi-visceral and vascular resection and reconstruction followed by a long course of anti-helminthics.

Here, report a case of a 49 year old female who presented with recurrence of hydatid disease after being operated for the same twenty years earlier

Case Report

In 2022, A 49 year old post menopausal women from India presented with complaints of vague continuous dull aching pain in the abdomen for two months. Patient had an episode of vomiting followed by fever with chills and rigor. She was conservatively managed for the same. There was no history of jaundice, dysuria or constipation. She had similar complaints twenty years previously and was diagnosed to have hydatid cyst and was operated for the same following which she had been asymptomatic till her presentation our outpatient department. She is a known hypertensive on regular medication. Informed consent was taken from the patient.

Ultrasonography revealed a hydatid cyst measuring 7.8 x 6.2 cm in segment IVa. Contrast CT revealed two well defined cystic lesions with peripherally arranged daughter cysts and central enhancing soft tissue areas in segment VII/VIII and IVa of liver measuring 12 x 10.6 x 9.8 cm and 9 x 8.6 x 6 cm. Cyst with similar picture was seen in the pouch of Douglas measuring 10.6 x 9.8 x 9 cm causing splaying and displacement of uterus to right side and left mild proximal hydronephrosis. Her liver function tests were within normal limits. She underwent exploratory laparotomy which revealed a cyst of 15 x 10 cm in the broad ligament of the uterus and another cyst occupying segment VII, VIII and IV of liver. Cystectomy was proceeded (Fig. 1 and 2).

Post operatively, she was managed with anti-helminthic albendazole and intravenous antibiotics.



Fig. 1: Postoperative images of the removed daughter cysts

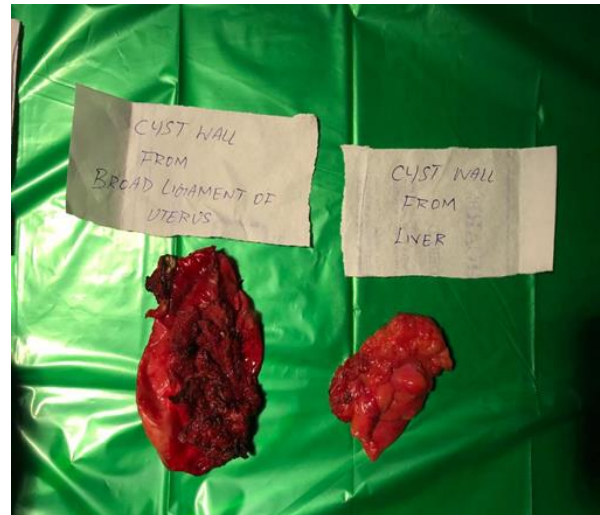


Fig. 2: Postoperative image of the removed cyst wall from the liver

On the third postoperative day, Magnetic Resonance Cholangiopancreatography was done which revealed a subdiaphragmatic collection of about 8 x 3 x 1.5 cm. Endoscopic Retrograde Cholangiopancreatography was

done which showed a leak from an intrahepatic biliary radical in the right lobe of liver with a subdiaphragmatic collection of contrast. 7.5 X 7 cm DP stenting was done in the right intrahepatic biliary radical system. Biliary sphincterotomy was done. The patient was on regular follow up for over a year and remained symptom free.

Discussion

Hydatid disease is a rare parasitic disease endemic in many parts of the world. It is a disease that is often misdiagnosed as the patients are either asymptomatic or present with vague abdominal symptoms. The clinical picture depends on the local effects of the parasite on the organ targeted, compressive symptoms or complications due to secondary infection, rupture and anaphylaxis. These complications are often fatal and is therefore one of the most dangerous helminthic diseases in humans.

Here, we are reporting the case of a 49 year old female who presented with recurrence of hydatid disease after being operated for the same twenty years earlier. The larval scolices of the parasite can mature into hydatid cysts in humans. Hence, operative spillage can result in regrowth of hydatid cysts after a suitable interval. The recurrence rate following surgery for hepatic hydatid disease is 10% (7). The surgery for a recurrent hydatid disease is challenging due to adhesions from the previous surgery and due to the spilt hydatid fluid. Our patient presented with concomitant cysts in the broad ligament. The main principle during surgery is to avoid intraperitoneal spillage of living cystic content (8). They remain in the watershed areas of the peritoneal cavity: The ileocolic region, the root of sigmoid and the Pouch of Douglas. These areas are to be carefully investigated in a patient presenting with suspected recurrence of hydatid disease. The other less plausible causes are primary infection through the alimentary tract or through the blood secondary to that of the liver.

The follow up period for hydatid disease should not be shorter than 3 years (9). Our case suggests that a patient with hydatid disease should be followed up for as long as possible and our patient presented with recurrence twenty years later. Post-operative medical therapy also influences the recurrence rates. In a study, regardless of the surgical approach, recurrence rate was 6.7% and 23.3% in patients who were given perioperative medical therapy and in patients who were not, respectively (10). Our case also serves to illustrate the importance of post-operative ERCP even in patients with no evidence of cystobiliary communication. Preoperative ERCP is to be considered in patients with cholangitis, jaundice and right upper quadrant pain. Post-operative ERCP is commonly indicated in patients with persistent drainage and obstructive jaundice with or without cholangitis. We conclude that *E. granulosus* can affect any organ with a propensity for unusual locations. A high index of suspicion is to be maintained while evaluating patients from endemic areas presenting with vague abdominal symptoms. A careful history has to be elicited from patients to rule out recurrence and meticulously evaluate areas that are susceptible to peritoneal spillage from previous surgery.

Acknowledgements

No financial support was received.

Conflict of Interest

None declared.

References

1. Akyildiz HY, Akcan A, Karahan I, Kucuk C, Sozuer E, Esin H. Recurrent liver hydatid disease: when does it become symptomatic and how does one diagnose it? Clin Imaging. 2009; 33(1):55-8.

2. Abdullah A, Alsafi R, Iqbal J, Rotimi V. Unusual case of pelvic hydatid cyst of broad ligament mimicking an ovarian tumour. *JMM Case Rep.* 2016; 3(4): e005057.
3. Goja S, Saha SK, Yadav SK, Tiwari A, Soin AS. Surgical approaches to hepatic hydatidosis ranging from partial cystectomy to liver transplantation. *Ann Hepatobiliary Pancreat Surg.* 2018; 22(3):208-15.
4. Salamone G, Licari L, Randisi B, et al. Uncommon localizations of hydatid cyst. Review of literature. *G Chir.* 2016;37(4): 180-185.
5. Bickers WM. Hydatid disease of the female pelvis. *Am J Obstet Gynaecol.* 1970;107: 477-483.
6. Rahman MS, Rahman J, Lysikewicz A. Obstetric and gynaecological presentations of hydatid disease. *Br J Obstet Gynaecol.* 1982;89: 665-670.
7. Sielaff TD, Taylor B, Langer B. Recurrence of hydatid disease. *World J Surg* 2001;25:83-86.
8. Bailey, Love. Norman S. Editors: Williams, Patrick Ronan, O'Connell AW. McCaskie. *Short Practise of Surgery.* 27th ed.
9. JM Little, MJ Hollands, Henrik Ekberg. Recurrence of Hydatid Disease. *World J Surg.* 1988;12(5):700-703.
10. Gallackner B, Langle F, Auer H, et al. Radical surgical therapy of abdominal cystic hydatid disease: Factors of recurrence. *World J Surg.* 2000;24:717-727.