

Case Report

Extensive pelvic hydatid disease mimicking ovarian malignant tumour

Rema Nair Sarkar¹, I. Vani², A. Bhagya Lakshmi^{1*}, T. Radha²

¹Department of Pathology, Andhra Medical College, Visakhapatnam, Andhra Pradesh, India

²Department of Gynaecology & Obstetrics, Andhra Medical College, Visakhapatnam, Andhra Pradesh, India

Received: 01 February 2015

Accepted: 19 February 2015

*Correspondence:

Dr. A. Bhagya Lakshmi,

E-mail: dr.a.bhagyalaxmi@gmail.com

Copyright: © the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

Hydatid disease is one of the commonest parasitic infections of the liver, endemic in many countries. Rupture into the peritoneal cavity leading to secondary echinococcosis is a difficult problem to manage. A case of 37 year old female patient presenting with disseminated intra-abdominal hydatid disease mimicking malignant multilocular cystic tumor of the ovary involving the mesentery of the small intestine, omentum and spleen is presented along with a brief review of literature.

Keywords: Multilocular ovarian cyst, Pelvic hydatidosis, Splenic cyst

INTRODUCTION

Echinococcosis is a multisystem disease and can mimic any disease process and has tendency to involve any organ. Peritoneal echinococcosis is generally secondary to a rupture of a liver-located hydatid cyst. Occasional cases of primary peritoneal hydatid disease including pelvic involvement have also been reported which is generally rare. Primary ovarian hydatid cyst is rarer. We report here pelvic hydatidosis mimicking a malignant multilocular ovarian tumor with no involvement of liver. Our patient, a 37 year-old female was detected to have a large right cystic adnexal mass on per vaginal examination which was confirmed by ultrasonography and MRI.

CASE REPORT

A 37 year-old female patient was admitted to gynaecology department with complaints of persistent dull aching pain in lower abdomen with distension of the abdomen and breathlessness of three months. Her past menstrual history was normal. There was no alteration of

bladder or bowel habits. Clinical examination revealed a large smooth surfaced mass (24×20 cm) arising from the pelvis with restricted mobility separate from the Uterus. Adnexae could not be palpated on P/V examination. Ultrasound (USG) of the whole abdomen revealed a huge right ovarian 57 mm × 70 mm heterogeneous semi-solid multiloculated cystic mass with multiple cysts of varying sizes in the pelvis and peritoneum. The right ovary was not visualized. Left ovary was normal. Ultrasound did not detect free fluid within the abdomen. MRI showed multiple multiloculated, hyperintense cystic lesions of varying sizes located in subphrenic, subhepatic, bilateral paracolic gutters. Right ovarian growth was noted as 60 × 45 mm heterogeneously multiloculated cystic mass with septations. There was no evidence of free fluid in the peritoneal cavity but showed minimal right pleural effusion. Moderate splenomegaly with two thick walled multiloculated cystic lesions of 1.5 mm x 1.5 mm each were seen also seen in the spleen. The liver, kidneys and bladder showed a normal echo pattern. LFT and other lab investigations were within normal range. CA125 was mildly elevated. Depending on these indicators the provisional diagnosis of a possibility of a pelvic

hydatidosis or a right ovarian malignant cystic neoplasm with peritoneal metastasis was offered. Exploratory laparotomy was performed (Figure 1). Multiple cystic masses were seen arising from the pelvis incorporating the right adnexa, covering the peritoneum, mesentery of small intestine paracolic areas with widespread adhesions obliterating the pouch of Douglas. Uterus was pushed to left with left tube and left ovary appearing normal. The cystic masses were excised and total abdominal hysterectomy and bilateral salpingo-oophorectomy performed. Around 35-40 cystic masses of 2 cm to 10 cm diameter were excised without compromising the vascularity of the intestine with separate or along with omentum. Externally there were no palpable lesions on hepatic and splenic surfaces. Intraoperative findings were consistent with multiple hydatid cysts in viscera. The resected cysts were processed histopathologically.

Grossly we received total hysterectomy specimen with bilateral adnexa along with numerous white colored cyst with glistening surface, largest measuring 10 cm x 6 cm. (Figure 3). The right ovary could not be identified as it was incorporated and adhered to cysts of varying sizes.

On cut sections large cysts revealed numerous daughter cysts. Microscopic examination of fluid from the cysts revealed free scolices with hooklets (Figure 2) and the biopsy showed cyst wall with typical laminated membrane with inner germinal layer containing degenerated protoplasmic mass. The diagnosis of pelvic hydatid disease was confirmed (Figure 4) and patient was managed accordingly. Post-surgery was uneventful. The patient was given prophylactic hydrocortisone and antihistaminics for 24 hours with Albendazole 10 mg/per kg body weight for 6 weeks and discharged. She has been followed up for the last 4 months with repeat USG at each follow visit. As yet there is no evidence of any recurrence or increase in size of splenic cysts.

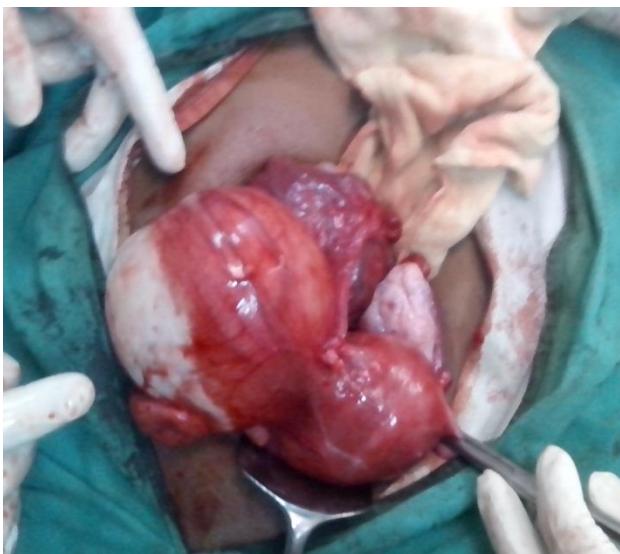


Figure 1: Intraoperative photo showing multiple hydatid cysts.

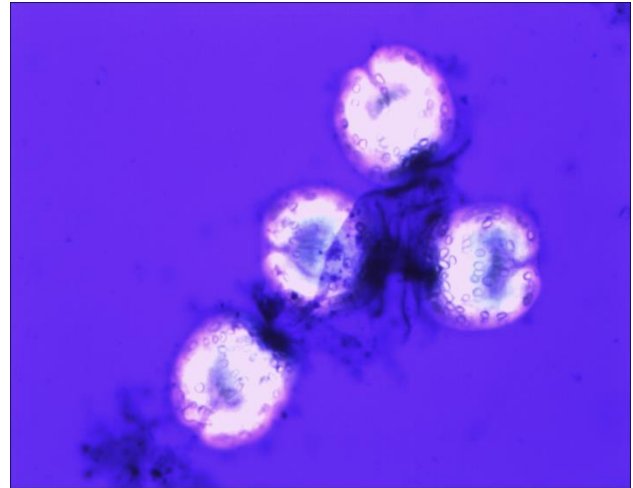


Figure 2: Hydatid scolices seen in the resected cyst fluid (40x toluidine stain).



Figure 3: Hydatid cysts and the uterus with the incorporated adnexa.

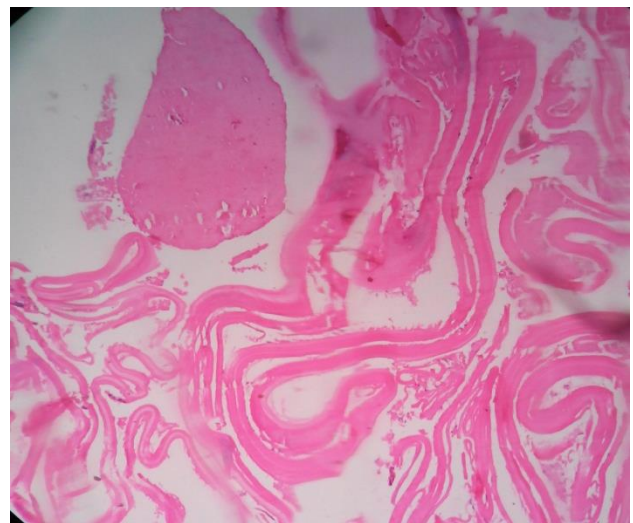


Figure 4: Lamellated wall of the hydatid cyst (100x H&E).

DISCUSSION

Hippocrates more than 2000 years ago described hydatid disease in humans first and used the term 'liver filled with water' by and subsequently a famous Arab physician Al-Rhazes mentioned about the disease about 1000 years ago.¹ Human infection occurs when the eggs of *Echinococcus* are ingested, either by consuming contaminated unwashed vegetables or as a result of close association with pet dogs.² Hydatid disease is prevalent in areas where livestock is raised in association with dogs. It is found mostly in countries where people live in close association with livestock. The organs most commonly involved in hydatid disease are the liver and lungs.⁴ The overall prevalence of peritoneal involvement in cases of abdominal hydatid disease is approximately 13%.³ Pain is the most common symptom of hydatid disease as present in our case. Fever supervenes in secondary infection and intraperitoneal rupture causes severe allergic reactions. Jaundice might develop in hepatic hydatid cysts when there is intrabiliary rupture. LFT was normal as our case had no cysts in liver [5]. Laboratory evaluation of patients with hydatid disease often yields non-specific data. A large battery of serological tests are available but their importance have been diminished by increased reliance upon modern imaging modalities like ultrasound (USG), Computed Tomography (CT) and Magnetic Resonance Imaging (MRI). A double contrast CT scan is 90-100% accurate for diagnosing hydatid cysts.⁶ This patient was diagnosed as a case with a possibility a hydatid cyst or a malignant cystic ovarian tumor on the basis of ultrasound CT and MRI investigations.

CA-125 considered gold standard tumor marker in evolution of pelvic masses particularly in ovarian epithelial cancers was mildly increased in our case probably due to extensive inflammation. This marker is elevated in 80% of ovarian epithelial cancers, 30% of non-ovarian malignancies, 6% of benign gynecological disorders, and 1% of normal cases. The benign conditions that cause elevation of CA-125 include pregnancy, ovarian cysts, uterine leiomyomas, pelvic inflammatory disease, and endometriosis. A preliminary diagnosis by either cytology or fine needle aspiration though contraindicated may not always be helpful as the thick mucin aspirated with poor cellularity may mimic the laminated membrane of hydatid and can easily be misinterpreted as ectocyst of hydatid disease.⁷ In our case the unusual presentation of pelvic hydatid disease with absence of ascitis, presence of multiple cysts in the pelvis, and a mildly positive serum marker (CA-125), it was very difficult to differentiate this from malignant cystic ovarian tumor with similar imaging findings.

The growth of hydatid cyst remains indolent, increasing their diameter by about two to three centimeters each year taking a latent period of five to twenty years depending not only on immunologic relationship between the parasites and humans but also on the resistance offered by the enveloping structure before the diagnosis is made.⁸

The outermost layer, the pericyst is extremely thin in peritoneal hydatid cyst unlike very thick walls in hepatic and splenic hydatid cysts due to fibrous tissue as a result of the reaction of the liver to the cyst. The innermost layer is the germinal epithelium (endocyst) which consists of a single layer cells lining the cyst, the living part of the hydatid cyst gives rise to Brood capsules. The release of brood's capsules, scolices and even daughter cysts from a ruptured hydatid cyst into the peritoneal cavity leads to multiple cysts in the peritoneal cavity.

The most common sites where hydatid cysts develop are liver (70%), which acts as a first filter and lungs (15%), which acts as second filter. The other 15% are found in other organs.⁹ In India the incidence of hydatid cysts at unusual sites is higher as compared with other parts of the world.¹⁰ Our case also had two splenic cysts which were present deep parenchymal and was left untouched during surgery on advice of the surgeons to be treated conservatively due to its location and size. The incidence of splenic involvement by hydatid cysts in relation to the rest of the abdominal viscera is very low.¹¹ Hydatid disease of spleen is extremely rare even in endemic areas (0.5-4 percent of all cases of echinococcosis).¹²⁻¹⁴ Splenic hydatid cysts have been reported coexistent with bilateral ovarian tumour.¹⁵ Mechanisms of primary pelvic hydatidosis are not clear. Genital organs are considered to be the most affected areas in the pelvis in females. This can be attributed to the fact that the genital organs are relatively highly vascularised, and other reasons could be invasion from the connective tissue of the peritoneum of Douglas and suspensory ligaments.^{16,17} Dissemination via lymphatics has been implicated as a possible route to produce primary pelvic hydatid disease.

The role of medical treatment in hydatid disease is limited with Surgery therefore being the mainstay of treatment for hydatid disease of the liver and is indicated in all patients with symptomatic disease. Following surgery, the reported recurrence rate is approximately 2% and survival rate is 95%.⁵

Recurrence rate after extra hepatic hydatid disease is very high as it is impossible to identify small residual seedlings. Only grossly visible disease can be removed. A postoperative long term follow up regimen is essential. Early postoperative imaging provides a baseline for late comparisons. Repeated imaging every 6 weeks is essential.

In conclusion, our case highlights that pelvic hydatid disease resembles malignant multicystic ovarian tumor, clinically and radiologically. The possibility of pelvic hydatid disease should be considered in endemic areas where differential diagnosis of cystic ovarian lesions is needed, so that the patient is managed accordingly. Hydatid disease must be considered while making the differential diagnosis of pelvic cystic masses, especially in endemic areas.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

REFERENCES

1. Katan YB. Intrabiliary rupture of hydatid cysts of liver. *Ann R Coll Surg Engl.* 1977;59:108-14.
2. Chatterjee KD. Hydatid disease. In: Chatterjee KD, eds. *Parasitology, Protozoology and Helminthology.* 13th ed. New Delhi, India: CBS; 2009: 159-165.
3. White AC Jr, Weller PF. Cestodes. In: Eugene B, Anthony SF, Dennis LK, Stephen LH, Dan LL, Larry J, eds. *Harrison's Principle of Internal Medicine.* 15th ed. USA: McGraw-Hill Companies, Inc.; 2001: 1248-1251.
4. Bickers WM. Hydatid disease of the female pelvis. *Am J Obstet and Gynecol.* 1970;107:477-83.
5. Mehra BR, Thawait AP, Gupta DO, Narang RR. Giant abdominal hydatid cyst masquerading as ovarian malignancy. *Singapore Med J.* 2007;48:e284-6.
6. Safioleas M, Misiakos E, Manti C, Katsikas D, Skalkeas G. Diagnosis evaluation and surgical management of hydatid disease of the liver. *World J Surg.* 1994;18:859-65.
7. Godara R, Dhingra A, Ahuja V, Garg P, Sen J. Primary peritoneal hydatidosis: clinically mimicking carcinoma of ovary. *Internet J Gynecol Obstet.* 2007;7(2):1-4.
8. Seymour I. Schwartz, Harold Ellis. Hydatid cyst. In: Seymour I. Schwartz, Harold Ellis, eds. *Maingot's Abdominal Operations.* 9th ed. New York: Appleton & Lange; 1989: 1226.
9. Singh KP, Sabhawal BD. Hydatid cyst of the breast. *Indian J Surg.* 1973;35:506-7.
10. Majumdar P, Ghosh DP. Hydatid cyst of the thyroid. *Indian J Surg.* 1970;32:496-9.
11. Davis CE, Montero JM, VanHorn CN. Large splenic cysts. *Ann Surg.* 1971;173:686-92.
12. Golematis B, Delikaris P. Treatment of echinococcal cyst. In: Golematis B, Delikaris P, eds. *Mastery of Surgery.* Boston, Massachusetts: Little Brown; 1984: 633-641.
13. Humphreys WG, Jhonston GW. Splenic cysts: a review of six cases. *Br J Surg.* 1979; 66 : 407-8.
14. Manouras AJ, Nikolaou CC, Katergiannakis VA, Apotolidis NS, Golematis BC. Spleen-sparing surgical treatment for echinococcosis of the spleen. *Br J Surg.* 1997;84:1162.
15. Thenevala SM, Chugh S. Splenic hydatidosis. *J Obstet Gynaecol India* 1963;13:146.
16. Gupta A, Kakkar A, Chadha M, Sathaye CB. A primary intrapelvic hydatid cyst presenting with foot drop and a gluteal swelling. *J Bone Joint Surg Br.* 1998;80:1037-9.
17. Terek MC, Ayan C, Ulukus M, Zekiopglu O, Ozkinay E, Erhan Y. Primary pelvic hydatid disease. *Arch Gynecol Obstet.* 2000;264:93-6.

DOI: 10.5455/2320-6012.ijrms20150433

Cite this article as: Sarkar RN, Vani I, Bhagya Lakshmi A, Radha T. Extensive pelvic hydatid disease mimicking ovarian malignant tumour. *Int J Res Med Sci* 2015;3:980-3.