Pelvic Hydatidosis Mimicking a Malignant Multicystic Ovarian Tumor

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Abstract: Echinococcosis is a multisystem disease and has propensity to involve any organ, an unusual anatomical site, and can mimic any disease process. Primary peritoneal echinococcosis is known to occur secondary to hepatic involvement but occasional cases of primary peritoneal hydatid disease including pelvic involvement have also been reported. We report here 1 such case of primary pelvic hydatidosis mimicking a malignant multicystic ovarian tumor where there was no evidence of involvement of the liver or spleen. Our patient, a 27-year-old female, was detected to have a large right cystic adnexal mass on per vaginal examination which was confirmed by ultrasonography. Her biochemical parameters were normal and CA-125 levels, though mildly raised, were below the cut off point. She underwent surgery and on exploratory laparotomy, another cystic mass was found attached to the mesentery of the small gut. The resected cysts were processed histopathologically. On cut sections both large cysts revealed numerous daughter cysts. Microscopic examination of fluid from the cysts revealed free scolices with hooklets and the cyst wall had a typical laminated membrane with inner germinal layer containing degenerated protoplasmic mass. The diagnosis of pelvic hydatid disease was confirmed and patient was managed accordingly. Hydatid disease must be considered while making the differential diagnosis of pelvic cystic masses, especially in endemic areas.

Key words: Echinococcus granulosus, hydatidosis, daughter cysts, multicystic ovarian turnour

INTRODUCTION

Hydatid disease is a parasitic infection caused by the tapeworm, Echinococcus, most commonly Echinococcus granulosus. The disease is endemic in sheep and cattle grazing countries like India, Australia, Middle East, Africa, South America, and Turkey [1]. The dog is the definitive host, harbouring the adult tapeworms, and the sheep, pig, cattle, goat, and man are intermediate hosts, harbouring the larval stage [2]. Man gets infected either by close contact with the definitive host or by consuming vegetables contaminated with eggs of Echinococcus [3]. The most common organs involved by hydatid disease are the liver (75%) and lungs (15%). The spleen, ovaries, brain, bones, and heart are the other sites involved by the disease [4]. Mesenteric involvement by hydatid disease is very rare and this is the second case reported in the literature so far [5]. Hydatid disease in extrahepatic locations is usually asymptomatic but as the cyst grows it may produce pressure symptoms or allergic reactions and infections due to rupture [6]. We report a case of a large hydatid cyst involving the right ovary mimicking a malignant

CASE REPORT

A 27-year-old female reported to our institute with a history of pain in the abdomen, backache, and menorrhagia of 2 months duration. She had a past history of cholecystectomy done 5 years back. On examination patient had pallor and slightly distended lower abdomen. Per vaginal examination revealed a right cystic adnexal mass. Routine haematological parameters revealed mild anemia with a hemoglobin of 10.3 g%. Biochemical parameters, including the liver function and kidney function tests, were normal. Ultrasonography of the abdomen showed the uterus normal in size and echotexture, and a multicystic septate mass was seen in the right adnexa with solid areas measuring $11.0 \times 7.0 \times 3.0$ cm. The left adnexa were normal. Study of other organs was normal. Patient's CA-125 levels were mildly raised to 60-70 U/ml (reference range < 35 U/ml). Her X-ray chest was normal. Exploratory laparotomy was done in which a right sided large cystic mass measuring $10.0 \times 5.0 \times 3.0$ cm involving the right ovary and adherent to the fallopian tube was found. Another large cyst measuring $7.0 \times 6.5 \times 3.0$ cm adher-

multicystic tumor along with a large hydatid cyst involving the mesentery of the small gut.

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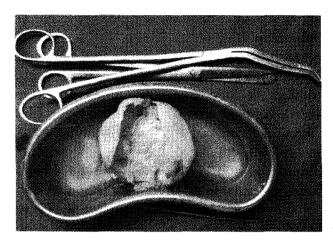


Fig. 1. A large hydatid cyst removed from the right ovary, showing numerous daughter cysts.

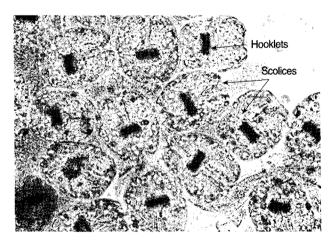


Fig. 2. Wet mount preparation from the hydatid fluid. A photomicrograph showing numerous scolices. × 200.

ent to the mesentery of the small gut was detected. Total abdominal hysterectomy with right salphingo-oopherectomy with mesenteric cyst removal was done.

At gross examinations, the cyst from the right ovary measured $10.5 \times 5.0 \times 3.5$ cm, multilocular, and contained many daughter cysts (Fig. 1). The cyst from the mesentery of the small gut measured $8.0 \times 6.0 \times 2.5$ cm. Cut section revealed numerous daughter cysts. The examination of the fluid aspirated from the cyst demonstrated free scolices showing hooklets (Fig. 2).

Histopathological examinations of the right adnexal mass showed some structure of the ovary containing the hydatid cyst. The cyst wall had a laminated membrane with inner germinal layer containing degenerated eosinophilic protoplasmic mass (Fig. 3). The mesenteric cyst revealed similar histopathological findings of the hydatid cyst.

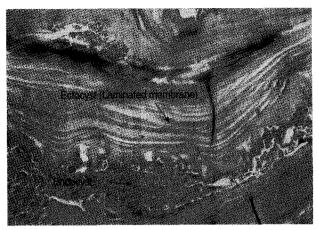


Fig. 3. A photomicrograph showing an ectocyst and endocyst. H- E stain. \times 400.

DISCUSSION

Human hydatid disease was described by Hippocrates more than 2,000 years ago who used the term 'liver filled with water' and subsequently about 1,000 years ago a famous Arab physician Al-Rhazes mentioned about the disease [7]. The disease is caused by the parasitic tapeworm species, *E. granulosus*, in the majority of the cases. 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]. Whereas the liver and lung hydatids are the most common type, peritoneal hydatid remains an uncommon manifestation [1]. However, secondary peritoneal involvement may occur through transcoelomic spread from rupture of the hydatid cyst of the liver or rarely spleen [6]. Spillage of the cystic fluid during surgery is considered another possibility [1].

Primary peritoneal hydatosis is rare, and has been reported to occur only in 2% of all abdominal hydatid disease cases [1]. Primary ovarian hydatid disease is also very rare with only very few cases reported in the literature [8]. There are many similarities between the hydatid cyst and other pelvic malignant diseases on the basis of imaging findings. Daughter cysts may resemble septal structures and mimic multicystic ovarian malignancy like cystadenocarcinoma. Moreover, in endemic areas where the hydatid disease is prevalent, it should be considered among the differential diagnosis of cystic pelvic masses [9].

In our case, the picture was different, where there was no evidence of hydatid disease of the liver or lung in the X-ray of the chest, ultrasonography, or CT scan. The patient had undergone cholecystectomy 5 years back, even then there was no recorded

evidence of hydatid disease of the liver or peritoneum. The patient was diagnosed as a case of malignant cystic ovarian tumor on the basis of radiological investigations. Even serologically the patient had raised levels of CA-125 marginally which could be explained on the basis of being reactive in nature and more so the levels returned back to normal after the surgery. CA-125 is a gold standard tumor marker in evolution of pelvic masses particularly in ovarian epithelial cancers. 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. CA-125 levels are commonly below 35 IU/ml and cut off point from I65-200 IU/ml is necessary to distinguish benign conditions from malignant lesions.

In a situation like ours, 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 is very difficult to differentiate this from malignant cystic ovarian tumor with similar imaging findings. A preliminary diagnosis by either cytology or fine needle aspiration 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 [10].

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 [11,12]. Dissemination via lymphatics has been implicated as a possible route to produce primary pelvic hydatid disease [13].

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 included in endemic areas where differential diagnosis of cystic ovarian lesions is needed, so that the patient is managed accordingly.

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