

Quiz

Hydatid disease: A parasitic inundation

Bahadur et al. Hydatidosis with Endo-myometrial involvement

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What is your diagnosis?

A 35year old lady, para 4 with 3 living issues and farmer by occupation presented to our hospital with complaint of acute urinary retention for which an indwelling urinary catheter was inserted. There were no associated complaints of haematuria, frequency, urgency or burning micturition. She also had history of progressively increasing lump abdomen for past 6 months with dull aching pain. Patient gave history of having undergone laparotomies twice during her childhood for a liver pathology but she was unaware of actual diagnosis. No records were available.

Physical examination revealed a 10x8cm cystic abdominal mass in left lumbar region. The mass was freely mobile and non-tender. There was another large abdomino-pelvic lump corresponding to 18-20 weeks period of gestation, cystic, mobile from side to side, non-tender and lower border could not be made out. Per vaginal examination revealed a normal sized uterus with a large 10x15cm cystic mass felt through right and posterior fornix.

Ultrasound revealed two large multiseptated cystic lesions on right and left side reaching up to epigastrium with multiple small cysts seen in both masses with no increase in vascularity, no solid areas and no free fluid in abdomen (Figure 1).

Contrast enhanced MRI showed non-enhancing peritoneal and omental based clusters of non-loculated cystic lesion in mid abdomen 14x11x11cm, in left iliac fossa 6x7x7cm and in pelvis 13x10x10cm with multiple small cysts within. (Figure 2).

Patient was planned for a laparotomy. Intra-operatively, uterus was normal size. A 10x15 cm clear cyst was seen arising from left ovary with multiple small cysts with viscous pale, yellow fluid inside them. Similar 2x2cm cyst was seen arising from right ovary, 20x10cm cyst and a 5x6cm cyst was seen arising from greater omentum (Figure 3). Total abdominal hysterectomy with bilateral salpingo-oophorectomy with omental cyst excision was done.

Answer

Histopathological examination revealed hydatid cysts lined by innermost germinal layer with attached brood capsule and daughter cysts. Avascular, eosinophilic and refractile laminated membrane was seen. Outer layer was fibrovascular with chronic inflammatory cells and giant cell reaction. To our surprise, even the myometrium had evidence of hydatid cysts (Figure 3)

The patient was a farmer and livestock handler by occupation. Ultrasound and MRI findings along with history of two previous surgeries in past raised a high index of suspicion for hydatid cyst. Pre-operative echinococcal serology was positive. CA 125 was normal. With a provisional diagnosis of suspected peritoneal and ovarian hydatid cyst patient was given a course of Albendazole 400mg twice daily for 6 weeks following which she was taken up for surgery that Echinococcus etiology of the cysts.

Echinococcosis or hydatidosis is caused by larvae of tapeworm *Echinococcus granulosus*, belonging to the family Taeniidae. Dog being the dominant carrier, Echinococcus often infects humans via echinococcus eggs in contaminated food, water or contact with infected animals (1). We present a case of a 35year old woman with co-existing ovarian, myometrial and omental hydatid cysts.

Hydatid disease is an endemic parasitosis in regions like China, Russia, South America, Mediterranean, Eastern Europe and Central Asia. (2) Prevalence of hydatid disease in liver or lung is 80%. (3) Very rarely the disease has pelvic or omental involvement. (4) Prevalence of pelvic hydatid cysts requiring surgery was reported as <1%. (5) Due to non-specific symptoms like abnormal uterine bleeding, sterility and urinary retention, diagnosis and treatment of pelvic hydatid cyst poses a challenge. Cysts with unusual location like ovary and uterus tends to grow slowly leaving the patient asymptomatic for a long time. (6)

The WHO Informal Working Group Classification of cystic echinococcosis(CE) is shown in Table 1 (3). Diagnosis can be made by imaging, USG or CT or MRI in conjunction with serology. USG lacks sensitivity for determination of cyst viability. The sensitivity of ultrasonography for evaluation of Echinococcus is 90 to 95 percent. (7) Amongst serological tests, specific IgG ELISA is the most sensitive measure. However, there is no consistent correlation between number or size of cysts and serologic results. (8) Serological tests are more reliable for diagnosis of *E. multilocularis* infection than *E. granulosus* infection. Antibody detection is more sensitive than antigen detection for diagnosis of *E. granulosus*. (9) Cyst aspiration –fluid PCR may also be useful for diagnosis.

Treatment modalities include PAIR (puncture-aspiration-injection-re aspiration) which is reserved for uncomplicated cysts that do not have daughter cysts (e.g. WHO stage CE1 and CE3a). (3) Percutaneous treatment is associated with risk of anaphylaxis. Modified catheterisation techniques are used to remove the entire endocyst and daughter cysts from cyst cavity using large bore catheters and cutting devices together with an aspiration apparatus. Drug therapy may be used for definitive or adjunctive therapy. Albendazole is the primary antiparasitic agent for treatment of *E. granulosus*. Surgery is the treatment of choice for complicated cysts or with cysts with many daughter vesicles (e.g. WHO stage CE2 and CE3b). Due to its high antigenic nature the toxic fluid of hydatid cyst cause anaphylaxis and recurrence. Hence cysts should be removed intact and if spillage occurs 15 to 20% hypertonic saline wash may be used. WHO recommends a postoperative chemotherapy with albendazole for 1 month or Mebendazole for 3months if spillage occurs (3). Since incidence of pelvic Hydatid cyst is very low and it mimics ovarian malignancy or other ovarian tumours, a high index of suspicion must be kept for this disease.

Our patient presented with cysts belonging to stages C2 and C3b where the preferred treatment is Albendazole followed by surgery which has been implemented for this patient. (3). Patient is healthy at a time of writing this case report having completed a 3 months course of albendazole postoperatively.

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Table 1. World Health Organization classification of cystic echinococcosis and treatment stratified by cyst stage

WHO stage	Description	Stage	Size	Preferred treatment	Alternate treatment
CE1	Unilocular anechoic cystic lesion with double line sign	Active	<5 cm	Albendazole alone	PAIR
			>5 cm	Albendazole + PAIR	PAIR
CE2	Multiseptated, "rosette-like" "honeycomb" cyst	Active	Any	Albendazole + either modified catheterization or surgery	Modified catheterization
CE3a	Cyst with detached membranes (water-lily sign)	Transitional	<5 cm	Albendazole alone	PAIR
			>5 cm	Albendazole + PAIR	PAIR
CE3b	Cyst with daughter cysts in solid matrix	Transitional	Any	Albendazole + either modified catheterization or surgery	Modified catheterization
CE4	Cyst with heterogenous hypoechoic/hyperechoic contents; no daughter cysts	Inactive	Any	Observation	-
CE5	Solid plus calcified wall	Inactive	Any	Observation	-

Albendazole is dosed 10 to 15 mg/kg per day in two divided doses; the usual dose for adults is 400 mg twice daily. Duration of therapy is discussed in the text; WHO: World Health Organization; CE: cystic echinococcosis; PAIR: puncture, aspiration, injection, reaspiration

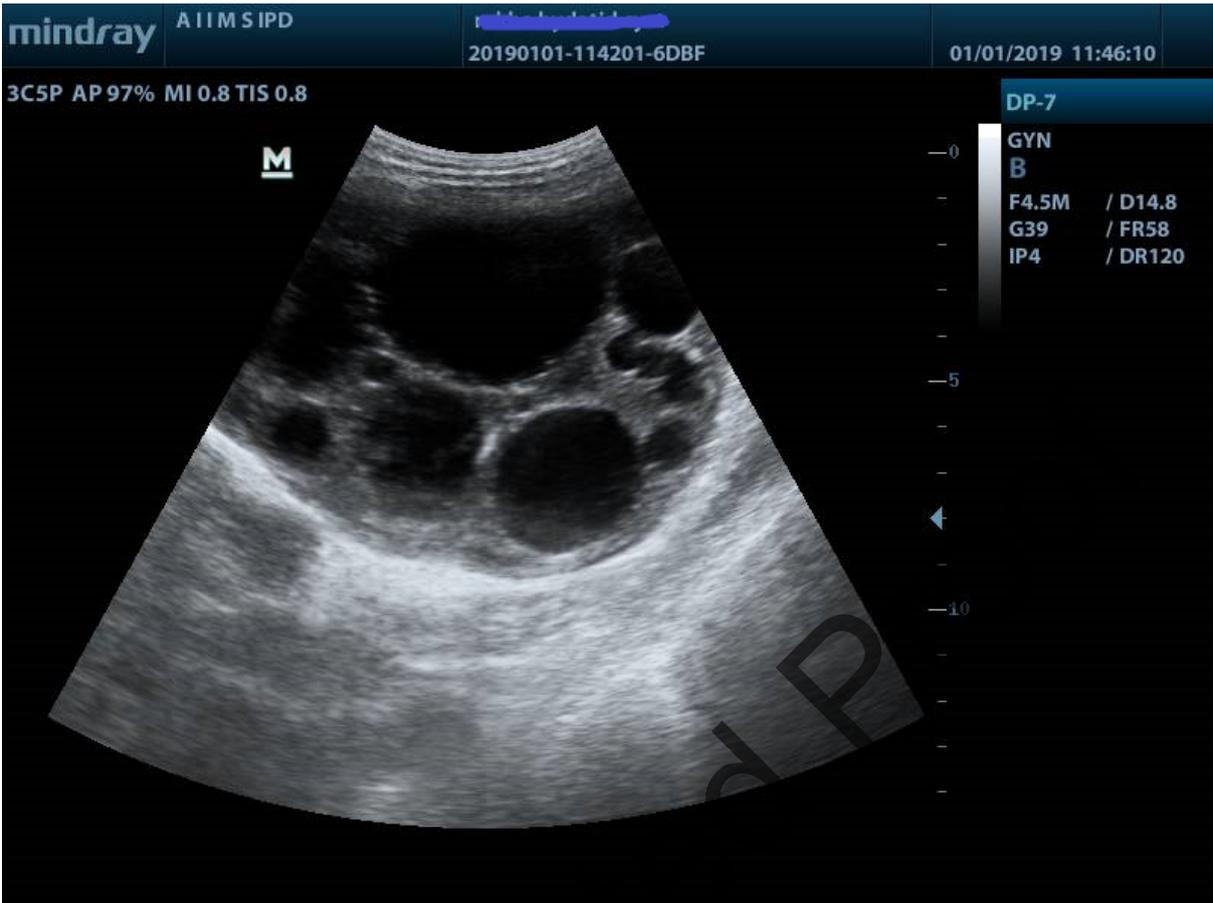


Figure 1: USG Pelvis showing multiseptated cystic lesion with multiple small cysts within in both adnexae

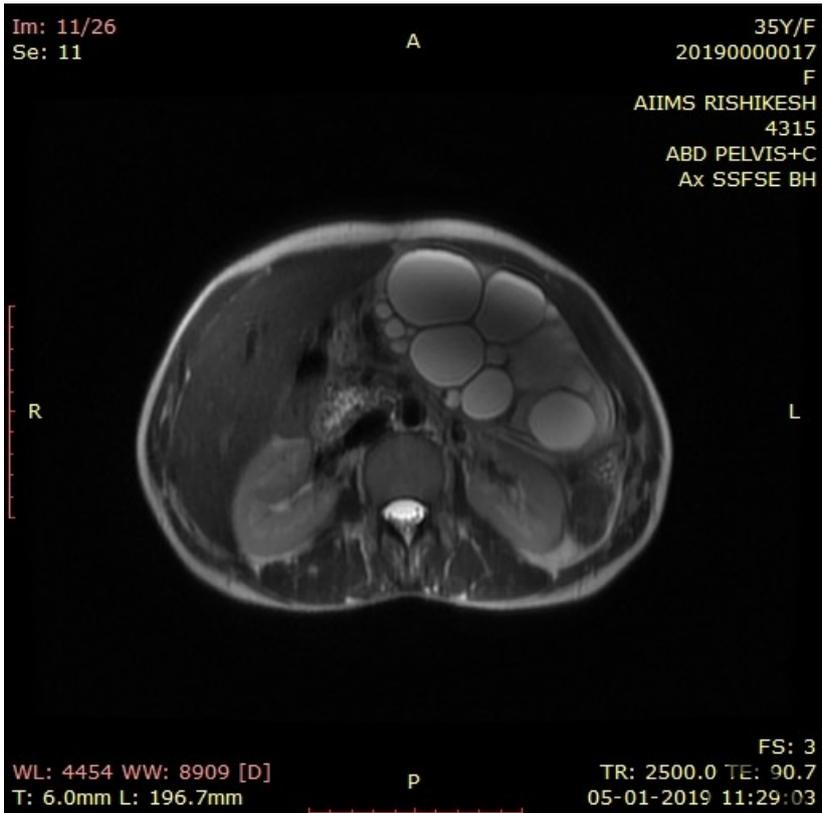


Figure 2: T2 weighted MRI image showing smooth walled clusters of fluid signal cystic lesion in pelvis with clusters of multiple tiny fluid signal cysts. Intra-operative finding showing multiple fluid filled cysts in ovary and omentum

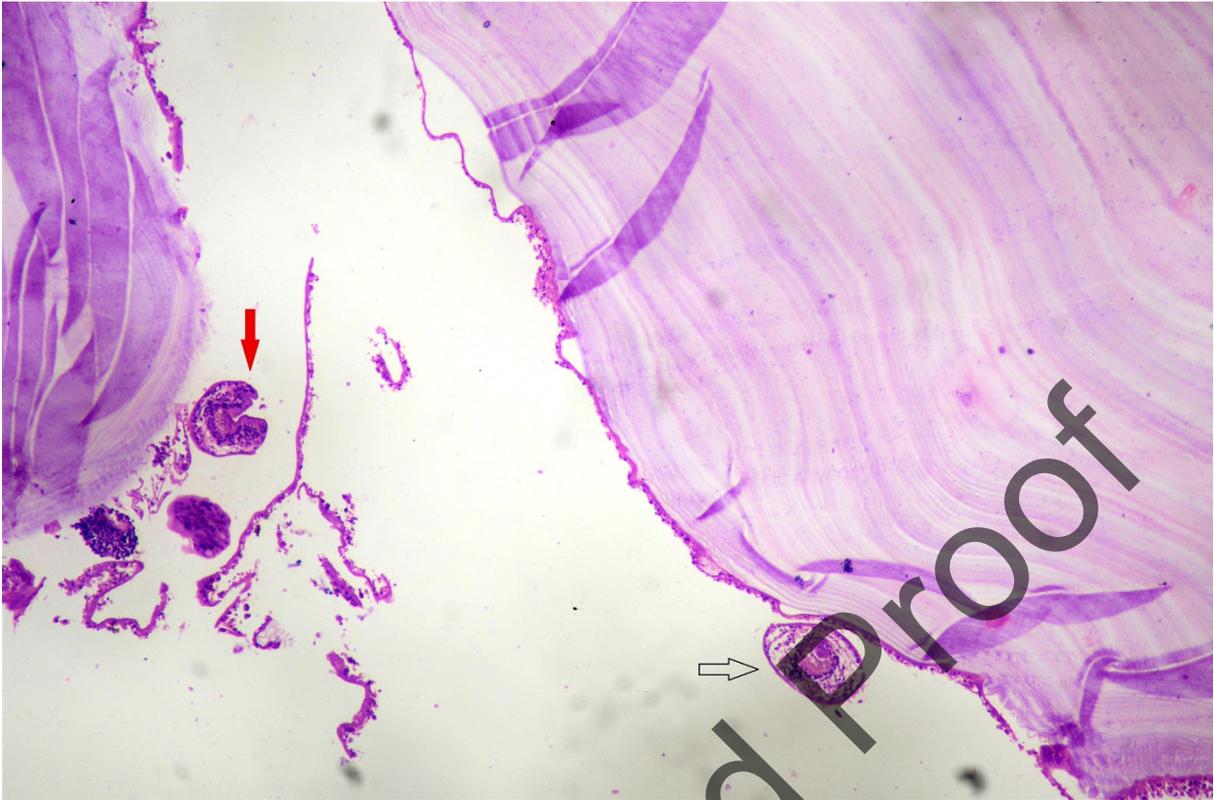


Figure 3: H&E (100X) Section shows lamellated membranes of *Echinococcus granulosus*. Also seen are brooding daughter cysts (black arrow) and scolex of the parasite (red arrow)