

## Hydatid Disease in Pregnancy: What to do and when to do?

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### Abstract

Echinococcosis also called as hydatid disease is a parasitic disease that affects various organs in the body. Due to its high incidence in India, we get to see a variety of uncommon disease presentations of which hydatid disease in pregnancy is one. As it of a rare occurrence, no management guidelines can be drawn. In this review we have summarized the available literature and tried to suggest some evidence based guidelines.

**Keywords:** Hydatid Disease; Pregnancy; Echinococcosis

### Introduction

Echinococcosis, also called as hydatid disease is a parasitic disease that affects various organs in the body. Hydatid disease is caused by *Echinococcus granulosus* and less commonly *Echinococcus multilocularis*. It is commonly acquired in childhood due to close contact with canines especially dogs (*Echinococcus granulosus* is also called dog tape worm). Humans accidentally swallow ova that are on their hands after touching dogs, and the embryo from the ova form cysts in their organs. Hydatid disease is endemic in Mediterranean countries, the Middle and Far East, South America, Southern Australia and East Africa. Due to its high incidence in India, we get to see a variety of uncommon disease presentations of which hydatid disease in pregnancy is one.

In general abdominal hydatid disease is asymptomatic unless complicated. Pregnancy can be accompanied by its own set of gastrointestinal symptoms and thus diagnosis of hydatid disease is impossible unless an ultrasonography is done. The incidence of hydatid disease in pregnancy is as low as 1 in 20,000 to 30,000 [1]. Due to its rarity no guidelines can be drawn. In this review we have summarized the available literature and tried to suggest some evidence based guidelines.

### Clinical features

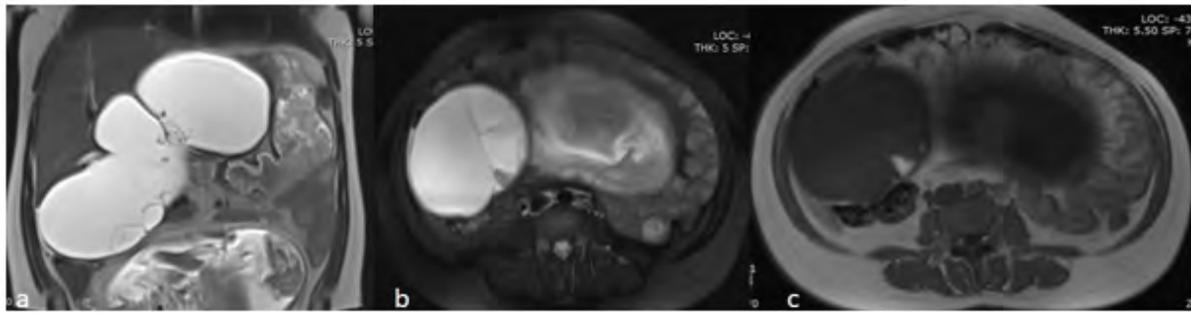
Hydatid disease does not have any specific clinical features and the same is true in pregnancy.

### The impact of pregnancy on hydatid and vice versa

Decreased cell-mediated immunity in pregnancy may facilitate rapid parasitic growth, and, as a result hydatid cysts may greatly enlarge in size leading to symptoms and complications [2,3]. Symptoms of mild epigastric pain due to reflux esophagitis may mimic a hepatic hydatid. Back pain in pregnancy mimics pelvic hydatid [4,5]. The enlarging uterus can cause rupture of hydatid and inevitable anaphylactic shock and dissemination of daughter cysts. Similarly hydatid cysts can result in uterine rupture during labour by contributing to the increased intra-abdominal pressure [6,7]. Pelvic hydatid cysts cause obstructed labour as well as premature labour [8]. Any surgical intervention in the later part of pregnancy increases risk of cyst rupture and premature labour.

## Diagnosis

**Imaging tests:** The best and easiest way to diagnose hydatid disease in pregnancy is an ultrasonography of abdomen. Single hydatid cyst can mimic simple hepatic cyst. Multi-vesicular cysts manifest as well defined fluid collections in a honeycomb pattern with multiple septa. Daughter cysts appear as cysts within a cyst. MRI has a complementary role to define the exact extent of the disease and for evaluation of internal characteristics. While ultrasonography is considered safe during all stages of pregnancy it has been concluded in a large cohort that exposure to MRI was not associated with increased risk of harm to the fetus. However gadolinium is associated with rheumatological, inflammatory, infiltrative skin conditions, stillbirth and neonatal death and should not be administered [9]. Computed tomography is best avoided during pregnancy due to risk of radiation to the fetus.



**Figure**

MRI abdomen and pelvis images of a 26 year old previously healthy patient who presented with severe abdominal pain at 28 weeks of gestation. Diagnosis was made on ultra-sonography complemented with serological tests and MRI was performed to define the disease extent. The patient was informed about the treatment options and side effects of each. She was advised surgery for which she did not give consent. She was put on oral albendazole. She went into spontaneous labour at 37 weeks and the second stage of labour was shortened by the application of forceps and corticosteroid cover was provided. A healthy baby was delivered and the post-partum course was uneventful and mild interval regression of the cyst was seen on follow up ultrasonography. She was again counselled for surgery for which she did not agree. Long term follow up of the baby's development is not available.

## Laboratory tests

Basic laboratory tests are nonspecific. They may indicate complications for example, an elevated total white count may indicate an infected hydatid cyst, elevated liver function tests especially alkaline phosphatase and gamma glutamyl transpeptidase may indicate biliary tract involvement with hydatid [10].

## Serological tests

These tests are based on the reaction and precipitation of test antigen and circulating antibodies in the host and include immunoelectrophoresis, enzyme linked immune-sorbent assay (ELISA) and western blot tests [11]. Immunoelectrophoresis has a diagnostic value of 91% to 94% for hepatic cysts and 69% to 70% for pulmonary cysts [12]. ELISA has sensitivity from 64% to 100% and western blot with purified antigens is highly useful [13,14]. The arc 5 antibody test has a specificity of 91% [15]. These tests are very useful for diagnosis and post treatment follow up.

**Treatment:** Can be divided into the following categories

- 1) Observation
- 2) Percutaneous aspiration
- 3) Chemotherapy
- 4) Surgery

The World Health Organisation (WHO) Informal Working Group on Echinococcosis described a US classification system which is intended to follow the natural history of hydatid disease [16]. This classification guides treatment of hydatid disease (Table 1).

Cyst Type	Status	Ultrasound Features	Remarks
CL	Active	Signs not pathognomonic, unilocular, no cyst wall	Usually early stage, not fertile; differential diagnosis is necessary
CE1	Active	Cyst wall, hydatid sand	Usually fertile
CE2	Active	Multivesicular, cyst wall, rosette like	Usually fertile
CE3	Transitional	Detached laminated membrane, water-lily sign, less round-decreased intracystic pressure	Starting to degenerate, may produce daughter cysts
CE4	Inactive	Heterogeneous hypoechogenic or hyperechogenic degenerative contents; no daughter cysts	Usually no living protoscoleces; differential diagnosis is necessary
CE5	Inactive	Thick calcified wall, calcification partial to complete; not pathognomonic but highly suggestive of diagnosis	Usually no living protoscoleces

**Table 1:** International classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings.

*CL: Cystic Lesions; CE: Cystic Echinococcosis.*

**Observation:** This is not a routine recommendation as cyst complications are more common during pregnancy which can be fatal for both mother and foetus. Observation is practised if the cysts are small and located in posterior liver segments, upper abdominal organs and retro peritoneum.

### Percutaneous aspiration

Percutaneous aspiration is popular technique for the treatment of hepatic hydatid.

Current indications for percutaneous treatment include:

- 1) Univesicular cysts: WHO classification cystic echinococcosis CE 1.
- 2) Univesicular cysts with detached membrane: WHO classification CE 3.
- 3) Some multi-vesicular cysts: WHO classification CE 2.

Contraindications include:

- 1) Cysts inaccessible to puncture.
- 2) Cysts in which puncture may damage structures in the vicinity.

Three different techniques of percutaneous treatment:

- 1) **PAIR:** Puncture, aspiration of the cyst contents, injection of protoscolicidal agents and reaspiration of cyst contents.
- 2) **PAIR catheterization technique:** After PAIR a catheter is introduced by Seldinger technique and the catheter is left in situ for drainage.
- 3) **PEVAC:** Percutaneous evacuation of cyst content, in which the PAIR-catheterization drainage is done initially and then the catheter is replaced by 14-18F stiff sheath. A suction catheter is introduced into the sheath and the contents are sucked. The cyst is continuously irrigated by protoscolicidals. Later the sheath is removed and catheter of the same size is placed. The catheter is removed subsequently once discharge is nil.

Percutaneous treatment should be considered in pregnancy as it is safe and is performed under sedation [17,18]. A meta-analysis comparing the clinical outcomes for 769 patients with hepatic hydatid cyst treated with PAIR plus albendazole or mebendazole with 952 controls undergoing surgery, Pair plus chemotherapy showed better clinical and parasitologic efficacy, lower rates of morbidity mortality, and disease recurrence; and shorter hospital stays [19]. Ustunsoz B., *et al.* studied 6 pregnant women with hepatic hydatid who underwent PAIR without albendazole therapy. They followed up the patients with sonography every 2 weeks during pregnancy to every 3 months post-partum in the first year, every 6 months in the second year and then yearly thereafter. First 5 cases developed solid appearance of the cyst indicating cure in 22 months. 6<sup>th</sup> case developed cystobiliary fistula which healed by conservative management [20]. Jayant., *et al.* described a case 32 weeks pregnant women with obstructive jaundice. On ultrasonography she has a Gharbi type II large hepatic hydatid cyst occupying segment V and VI. She underwent PAIR and was started on albendazole therapy. She delivered a normal baby. Post-partum albendazole was continued till 6 months. Ultrasonography done 3 months later did not showed any hydatid cyst [21].

### Chemotherapy

Chemotherapy consists of Albendazole. Albendazole cannot be given during the first trimester of pregnancy due to its teratogenic effects. Most common abnormalities detected include limb and facial abnormalities [22]. Treating hydatid cyst in pregnancy only with albendazole has been reported by Malhotra., *et al.* Malhotra., *et al.* reported a 22 weeks pregnant women who had hydatid cyst in left lobe of liver. She was treated with albendazole therapy till delivery. She had a full term normal delivery with a healthy baby. 2 months post-partum she underwent partial cystectomy. Van Vliet., *et al.* managed a 20 year old Turkish women detected with multiple hydatid cysts in liver only with albendazole and a cover of corticosteroids during labour [23].

### Surgery

Surgery in hydatid disease can be conservative or radical. Conservative techniques include deroofing of cysts and evacuation of daughter cysts. Radical techniques include cysto-pericystectomy and hepatic resection. Indication of surgery include large segment III and IV cysts and cysts opening into bile ducts and peritoneal cavity. The best time to perform surgery is in the second trimester; Though surgery has been performed during caesarean section as reported in 3 females in Libya [2]. One case of pelvic hydatid causing obstructed labour has been reported from India. She underwent caesarean section and intra-operatively the pelvic hydatid was excised completely [24]. Hemihepatectomy has also been performed during pregnancy [25].

The indication for surgery and the type of surgery should be individualised. Carlos Manterola reported 12 pregnant women with abdominal hydatid disease [26]. 11 had hepatic hydatid and 4 in addition had cysts in bladder, ovarian, peritoneum, mesentery and spleen. 9 of 12 patients underwent surgery. One in first trimester, 6 during second trimester and 2 in the third trimester and at delivery. The indications for surgery as per Carlos were hepatic cysts located in segment III, IV, V and VI, cysts in other abdominal organs which had grown into pelvis, cyst size more than 5 cms in diameter, cyst with imminent rupture, biliary tract communication, infection, and hepatothoracic transit.

### Summary and Conclusion

Any pregnant patient who complains of abdominal pain should undergo an ultrasound whole abdomen. Hydatid disease is difficult to diagnose in pregnancy due to overlapping symptoms. A high index of suspicion should be practised in endemic areas. WHO classification is a guide to treatment of hydatid cysts. Management of hydatid cyst includes observation, PAIR, chemotherapy and surgery. No guidelines are available for choosing the type of treatment. Observation is advisable in small hydatid cysts and retroperitoneal hydatid cysts though no specific size is mentioned. PAIR with or without albendazole is safe in pregnancy and is indicated in CE1, CE 3 and few CE 2 cases. Albendazole alone or with PAIR is advisable to be started only after first trimester due to its teratogenic effect. Surgery for hydatid cyst in pregnancy have specific indications. The safest period to operate is during the second trimester. Some surgeons have operated during the caesarean section. Emergency surgery may be needed in cases of intraperitoneal rupture, intrathoracic rupture, infection and pelvic hydatid causing obstructed labour.

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