

To Cite:

Patel NA, Acharya N, Borkar K, Patel T. Hepatic hydatidosis complicating pregnancy: A clinical challenge. *Medical Science* 2023; 27: e244ms2739.
doi: <https://doi.org/10.54905/disssi/v27i136/e244ms2739>

Authors' Affiliation:

¹Post Graduate Resident, Department of Obstetrics and Gynaecology, Datta Meghe Institute of Higher Education & Research, Sawangi, Wardha, Maharashtra 442005, India

²Professor, Department of Obstetrics and Gynaecology, Datta Meghe Institute of Higher Education & Research, Sawangi, Wardha, Maharashtra 442005, India

³Associate Professor, Department of Obstetrics and Gynaecology, Datta Meghe Institute of Higher Education & Research, Sawangi, Wardha, Maharashtra 442005, India

Peer-Review History

Received: 27 December 2022
Reviewed & Revised: 31/December/2022 to 20/May/2023
Accepted: 28 May 2023
Published: 2 June 2023

Peer-review Method

External peer-review was done through double-blind method.

Medical Science
pISSN 2321-7359; eISSN 2321-7367

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Hepatic hydatidosis complicating pregnancy: A clinical challenge

Nidhi Avinash Patel¹, Neema Acharya², Kiran Borkar³, Twisha Patel¹

ABSTRACT

Pregnancy is an immunologically vulnerable state. Due to various physiological and metabolic changes that take place during pregnancy, female body is more prone to pathogenic infections or precipitation of already established infection. Human hydatidosis is caused by *Echinococcus*, *Echinococcus granulosus* causes cystic echinococcosis in various organs of the body and *Echinococcus multilocularis* causes alveolar echinococcosis. Due to decreased cellular immunity and release of corticosteroids during pregnancy the asymptomatic hepatic hydatid cyst may enlarge in size and cause complications. Therefore, early diagnosis of hepatic hydatidosis and proper management plays a vital role in obtaining good fetal/maternal outcome.

Keywords: Pregnancy, Jaundice, *Echinococcus*, Hydatid cyst.

1. INTRODUCTION

Human hydatidosis i.e., hydatid cystic disease is a zoonotic parasitic disease caused by larval stages of tapeworms (cestodes) belonging to the genus *Echinococcus*. *Echinococcus granulosus* causes cystic echinococcosis in various organs and *Echinococcus multilocularis* causes alveolar echinococcosis. In its life cycle of accidental human intermediate host, the oncospheres invade the intestines, enter the vasculature where the liver acts as first filter for hydatid larvae making it the most common site to be affected, followed by lungs (Elshazly et al., 2009; Jain et al., 2022). Hepatic hydatid disease is generally asymptomatic for many years unless the cyst grows large enough to cause clinical signs and symptoms. Abdominal pain, nausea, vomiting, icterus are commonly seen with hepatic hydatid cyst. These are due to cyst compressing on the adjacent structures or rupture or infection of the cyst.

Hydatid cystic disease during pregnancy needs early diagnosis and treatment. Due to suppressed cellular immunity and release of corticosteroids by the placenta, the cyst growth is accelerated resulting in complications like compression of adjacent structures, rupture and communication with biliary tree. Moreover, the enlarging gravid uterus may compress on the hepatic hydatid cyst resulting in its rupture, spread of cystic fluid in the peritoneal cavity and anaphylactic shock (Sahin et al., 2005). Malpresentations, preterm

labour, dystocia are other complications associated with hydatid disease during pregnancy (Noori, 2021).

Hydatidosis is rare during pregnancy, but possess a life-threatening risk to both the mother and the foetus. Various imaging and serological tests are available for confirmation of diagnosis of hydatid disease during pregnancy. Treatment of hydatid disease include observational, medical, percutaneous aspiration and surgical. During pregnancy normal treatment is challenging due to risk of rupture, anaphylaxis, abortion and preterm labour. There is no standard or ideal treatment of hydatid disease in pregnancy (Ünalp et al., 2008). Here we present how we clinically diagnosed and managed a case of 23-year-old near term icteric pregnant female with abdominal swelling in right upper abdomen which on ultrasonographic examination was found to be hepatic hydatid cyst.

2. CASE PRESENTATION

A 23 years old primigravida with 8 months of amenorrhea, residing in rural area was brought to our tertiary rural hospital with history of swelling over right upper abdomen since 7 days. The swelling was sudden in onset, gradually increased in size, having regular margin and was associated with dull aching pain. There were on relieving or aggregating factors for pain and the pain did not radiate to any other site in the body. She gives history of yellowish discolouration of eyes since 4 days. There was no history of abdominal pain, fever, nausea, vomiting, constipation, pruritus, anorexia or breathlessness. The bowel and urine colour of the patient were normal. There was no history of hypertension, diabetes, thyroid disorder or bronchial asthma. She did not take any medications apart from routine antenatal medications. Obstetric examination on her first visit revealed primigravida with duration of marriage 7 years, her menstrual history revealed gestational age of 34 weeks 3 days.

On general examination patient’s vitals revealed pulse 98 bpm, blood pressure of 120/70 mmhg in right supine position and respiratory rate of 14/min. Conjunctiva of both eyes revealed icterus (Figure 1). There was no evidence of pallor, pitting edema or lymphadenopathy. On systemic examination cardiovascular and respiratory systems were normal. On per abdomen examination uterus was 32 weeks size with breech presentation and foetal heart rate was regular of 150 bpm. On per abdomen examination, inspection revealed a swelling in the epigastric and hypochondrium region of the abdomen. On palpation hepatomegaly was up to 8 cm below the costal margin with smooth surface. There was no evidence of local rise of temperature, tenderness or pulsatility over liver. No evidence of splenomegaly was found.



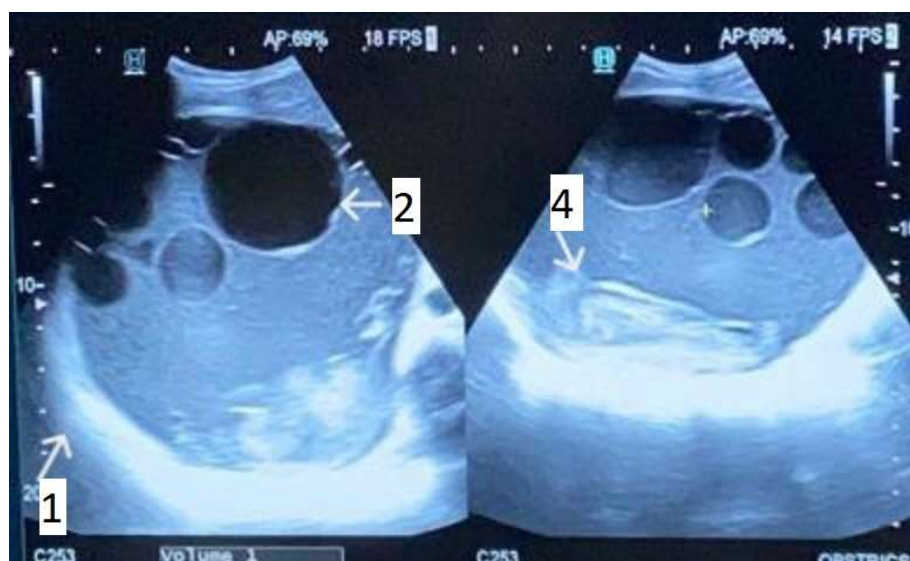
Figure 1 Conjunctival icterus seen in both the eyes of the patient

Table 1 Routine blood investigations

Laboratory Investigations		On admission	Post LSCS	2 weeks after pericystectomy
CBC	HB (gm/dl)	9.2	10.2	11
	MCV (fl)	72	74	75
	TLC (counts/dl)	13,400	9,000	10,200
	PLT (counts/dl)	1,10,000	1,45,000	1,00,000
Coagulation Profile	PT	13.3	13.4	13.3
	APTT	30.7	30.6	30.4
	INR	1.04	1.05	1.04
KFT	Urea (mg/dl)	12	11	10
	Creatinine (mg/dl)	0.3	0.4	0.3
	Sodium (mmol/dl)	135	133	135
	Potassium (mmol/dl)	3.8	3.5	3.4

LFT	Total protein (gm/dl)	5.4	5.2	5.1
	Albumin (gm/dl)	2.6	2.2	2.2
	Globulin (gm/dl)	3.1	3.3	3.1
	Aspartate aminotransferase (units/L)	58	22	22
	Alanine aminotransferase (units/L)	50	18	17
	Alkaline aminotransferase (IU/L)	124	90	90
	Total bilirubin (mg/dl)	3.3	2.3	0.9
	Conjugated bilirubin (mg/dl)	1.7	1.3	0.2
	Unconjugated bilirubin (mg/dl)	1.6	1	0.7

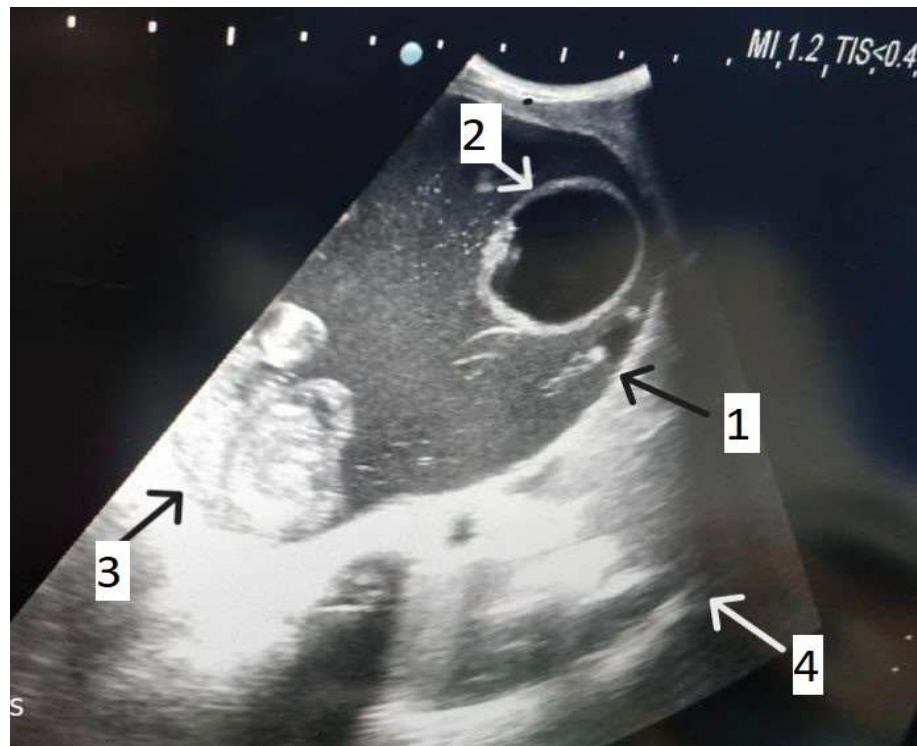
Routine laboratory investigations as in Table 1 were suggestive of elevated liver enzymes and elevated total, conjugated and unconjugated bilirubin. Serological tests for hepatitis B and hepatitis C were negative. Urine and stool analyses were normal. An antenatal ultrasound was performed (Figure 2a, 2b, 2c) which revealed single intrauterine live foetus of 32 weeks gestational age with 2 kg baby weight, longitudinal lie and breech presentation. Liquor was adequate and foetal Doppler was normal. There was evidence of large cystic lesion in right lobe of liver measuring 12 x 7 x 6 cm with multiple daughter cysts containing echogenic debris and crumpled membranes within it. MRI abdomen (Figure 3a, 3b, 3c, 3d) was performed and revealed large cystic lesion with multiple daughter cyst in the liver.



2a



2b



2c

Figure 2a, 2b and 2c Antenatal ultrasonographic images (1- Large hydatid cyst of size 12 x 7 x 6 cm, 2- Multiple daughter cysts, 3- Echogenic debris and crumpled membranes, 4- placenta and foetus)

A provisional diagnosis of near term primigravida pregnancy with hydatid cyst of liver was made. Foetal monitoring was done with DFMC and NST twice a day. 4 doses of Injection dexamethasone 6mg intramuscularly every 6 hours was given for foetal lung maturity. On consultation with gastroenterologist, tab ursodeoxycholic 300mg twice a day and tab albendazole 400mg twice a day was started for 21 days followed by 14 days no treatment interval, 3 such cycles are repeated. After 48 hours elective LSCS was performed and a pre-term alive baby of 2kg was delivered. Anti histaminics and steroids were given during LSCS.

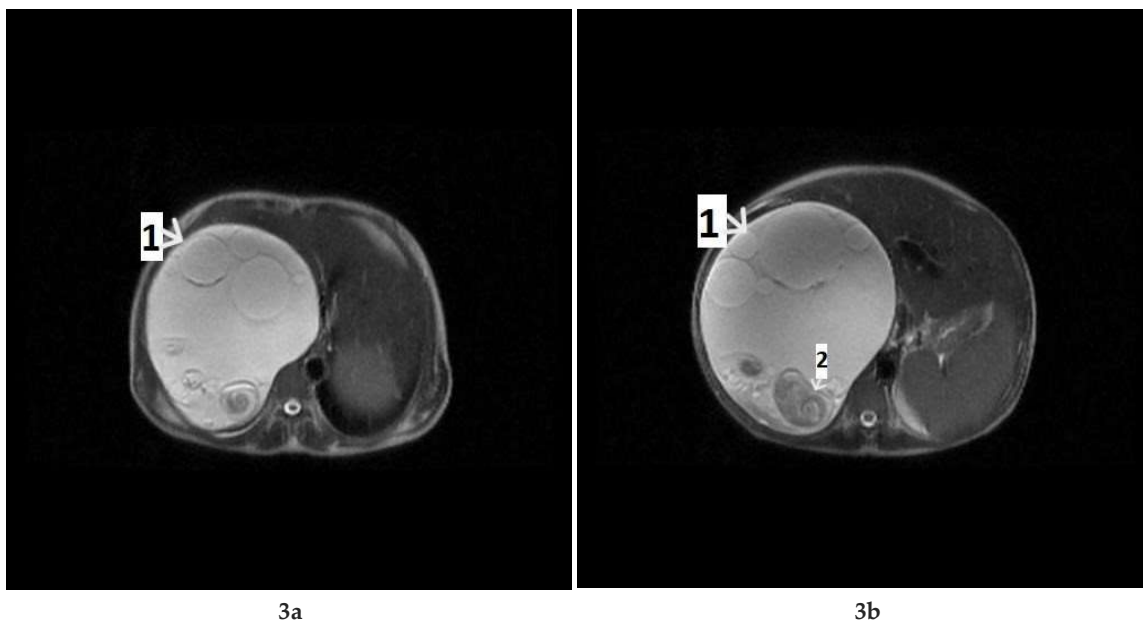


Figure 3a and 3b MRI images (axial planes)

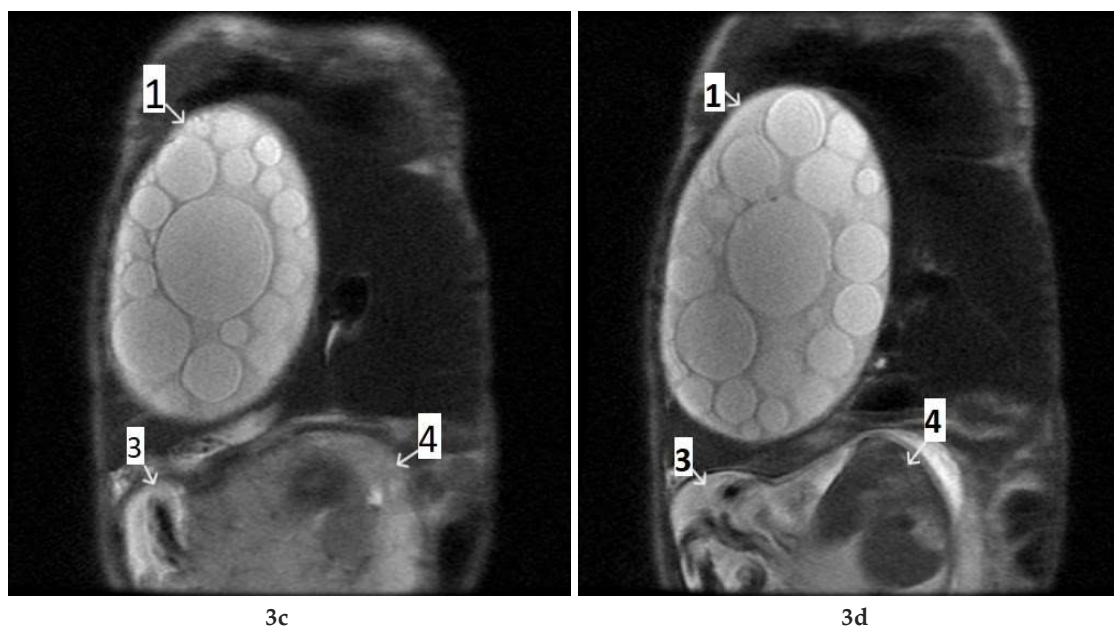


Figure 3c and 3d MRI images (coronal planes) (1- large cyst with multiple daughter cysts in V, VI, VII, VIII segments of liver, 2- debris and crumpled membranes, 3- placenta, 4- foetus.)

The baby and mother were discharged on day 7 of LSCS and was advised follow up to surgery opd after 3 months for pericystectomy of the hydatid cyst. She was discharged on Tab albendazole 400mg as per schedule described above. Patient was asked to continue ursodeoxycholic 300mg twice a day for one month.

Her routine laboratory investigations repeated on discharge post LSCS (Table 1). The elevated liver enzymes and bilirubin on admission were decreased on discharge. Two months post-partum a CT scan was performed to evaluate the size of hydatid cyst (Figure 5). Pericystectomy was performed by open method and samples sent for histopathology.



4a



4b

Figure 4a and 4b Abdomen on inspection during LSCS (1- Right hypochondrium swelling, 2- Gravid uterus)

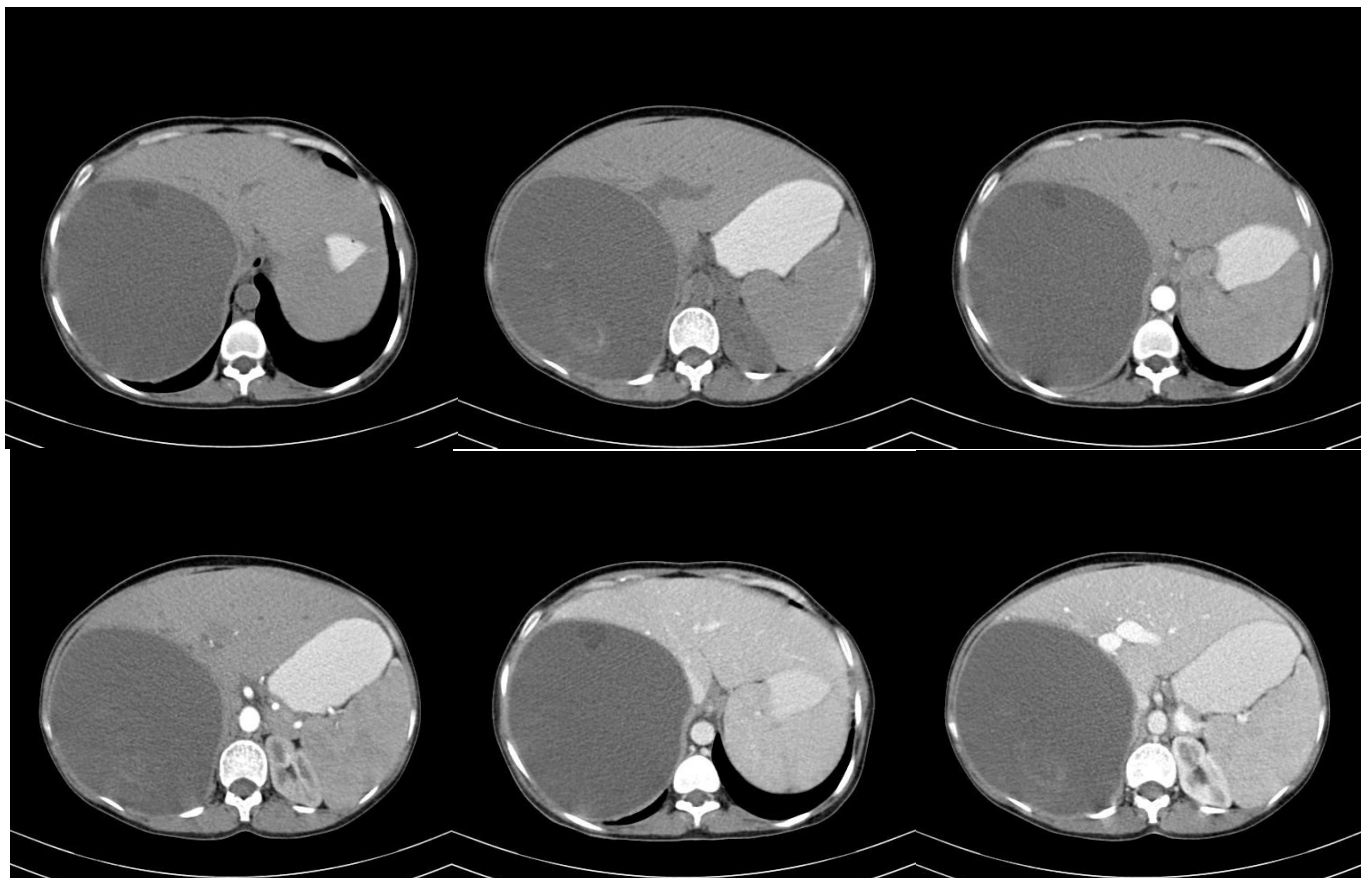
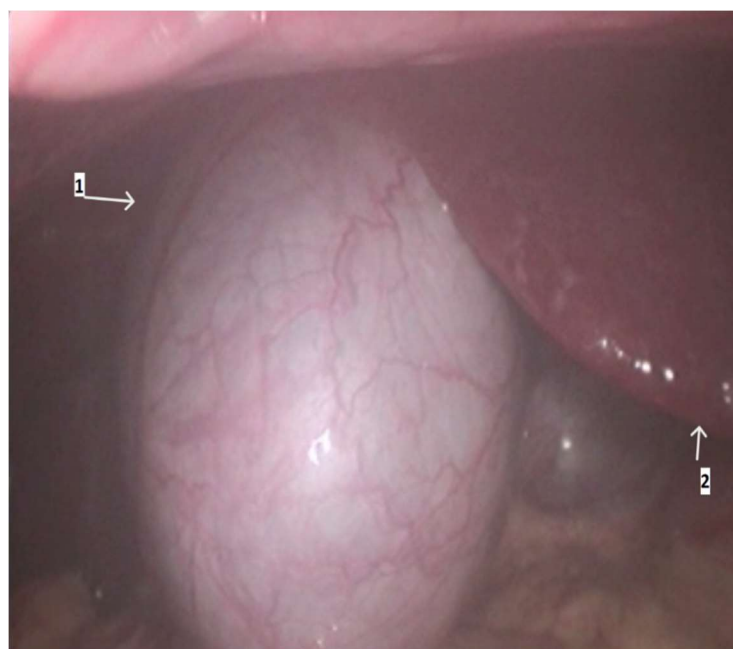
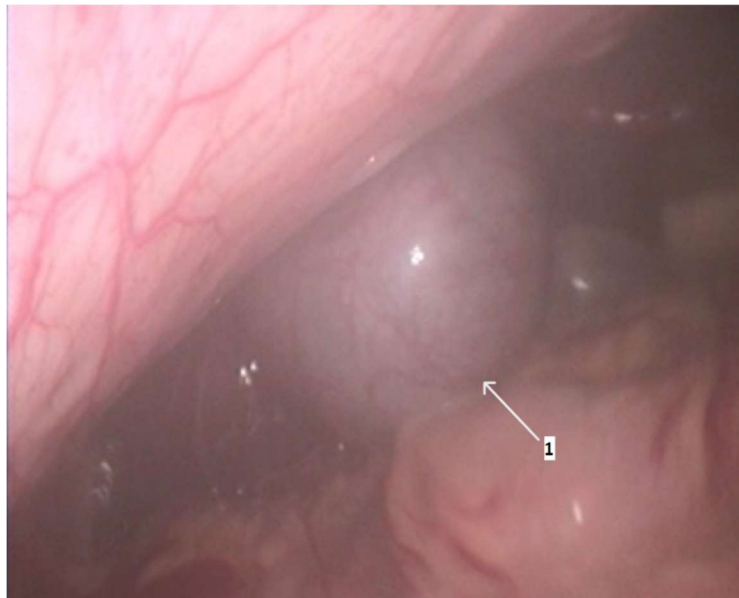


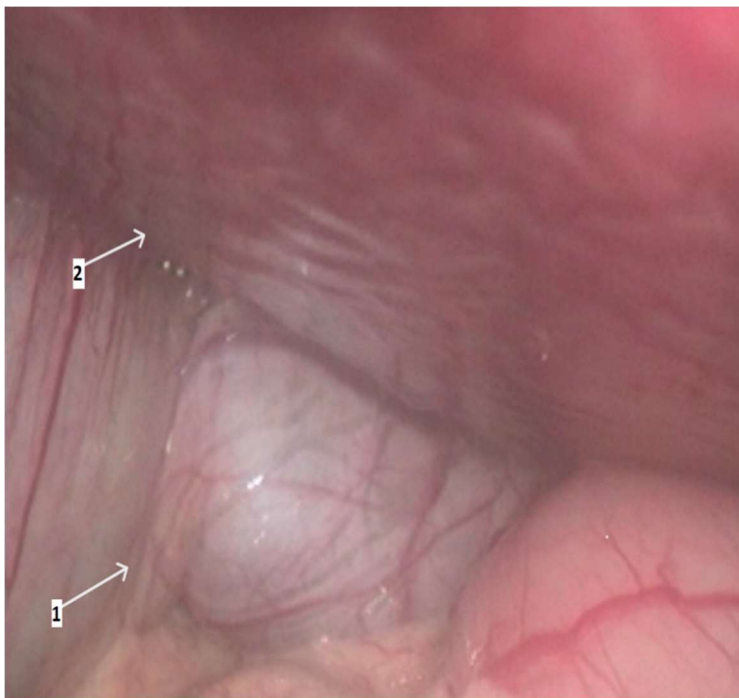
Figure 5 CT sections 2 months post-partum showing large cystic lesion in liver



6a



6b



6c

Figure 6a, 6b and 6c Intraoperative images during pericystectomy showing large hydatid cyst below right lobe of liver. (1- Hydatid cyst of liver, 2- Right lobe of liver)

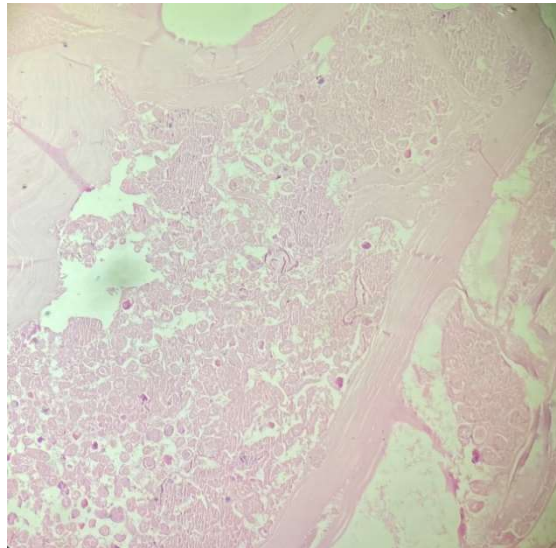


Figure 7a Section showing cyst wall and presence of brooding scolices within the wall

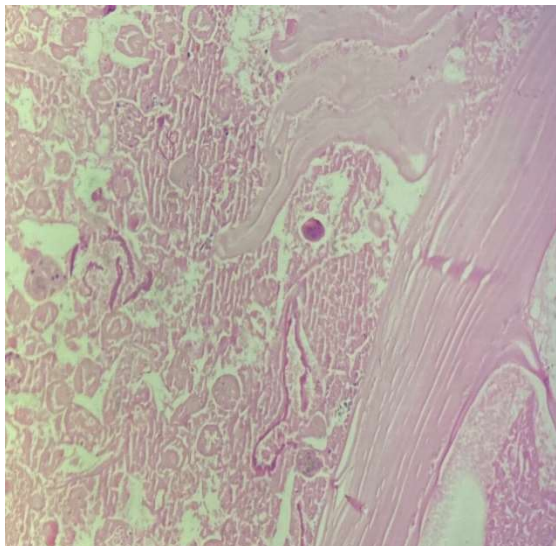


Figure 7b Section showing cyst wall lined by a laminated acellular membrane. Many ovoid shaped protoscolices can be seen

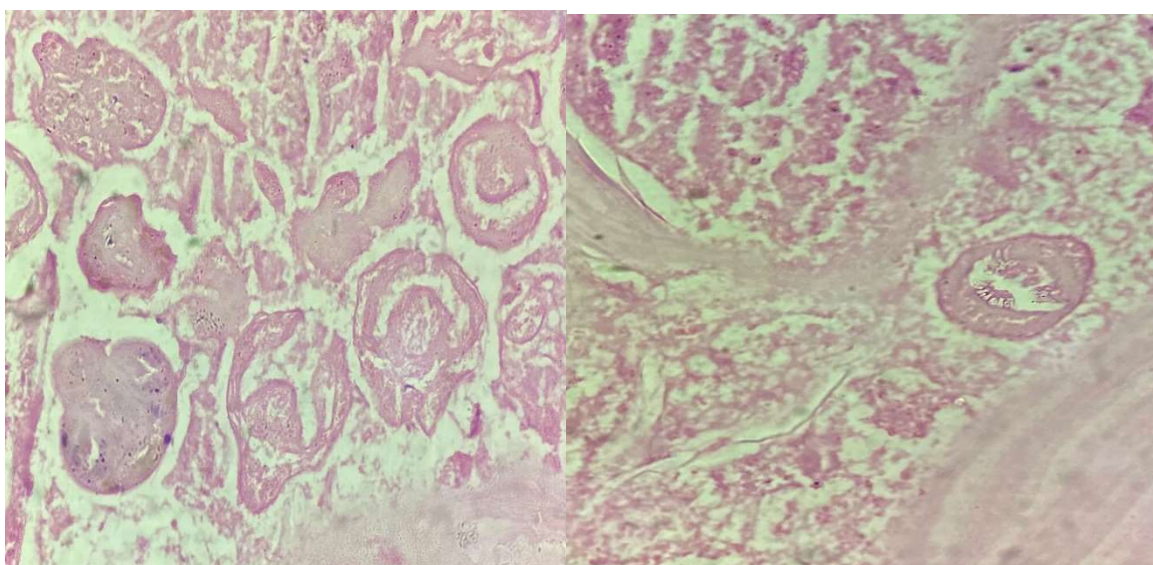


Figure 7c Section showing protoscolices with hooklets and sucker

Figure 7a, 7b, 7c- histopathology sections of hydatid cyst. Her routine laboratory investigations repeated 2 weeks after pericystectomy are as mentioned in Table 1 above. Post pericystectomy after 2 weeks routine investigations revealed normal liver enzymes and bilirubin levels.

3. DISCUSSION

Hydatid disease is a zoonotic disease caused by *Echinococcus granulosus* which mainly affects liver and lungs. The mature tapeworm is present in the intestine of carnivores animals (mostly dogs), the definitive host. Definitive hosts are infected by consumption of viscera of intermediate hosts (which contain larval parasite). Herbivorous and omnivorous animals act as intermediate hosts and are infected by ingesting food and water contaminated with parasite eggs. The parasite then develops into larval stages in the viscera of these intermediate hosts. Humans act as accidental intermediate hosts i.e., they acquire infection as other intermediate hosts but are not involved in transmitting the infection to definitive hosts.

Hepatic hydatid disease has a wide and variable spectrum of clinical symptoms and signs. Uncomplicated cases may remain asymptomatic for years according to size and extent of cyst. Complicated cases mostly present with right upper quadrant pain. Other symptoms include nausea, dyspnea and dysphagia. Hepatomegaly- a major sign of hepatic hydatid disease is demonstrated on clinical examination. Jaundice is commonly seen due to local compressing effect of enlarging cystic mass into the biliary tree. Due to secondary infection or rupture of cysts anaphylactic shock is common and is associated with fever and chills (Pedrosa et al., 2000; Dziri, 2001; Munzer, 1991; Ray and Deepak, 2017).

Hydatid disease which is rare during pregnancy ranging from 1 case per 20,000-30,000 population, can lead to serious maternal and foetal complications if not diagnosed and treated early. The cyst may remain small for many years and grow rapidly during pregnancy due to immunocompromised state as a result of decreased cellular immunity and release of corticosteroids by the placenta. They may present clinically as described above. Various complications during pregnancy include malpresentations, intrauterine growth restriction, pre-term labour, dystocia, anaphylactic shock, etc. (Noori, 2021). Radiological modalities like ultrasonography, MRI, CT scan can be used to detect and confirm hydatid cyst. Ultrasonography (USG) is used to detect cystic membranes, septa and hydatid sand. Computed tomography (CT) and magnetic resonance imaging (MRI) is used to demonstrate cystic wall defects and passage of contents through the cyst wall (Pedrosa et al., 2000).

Ultrasonographic classification of hydatid cyst is as follows (Noori, 2021)

Ghabri classification	Who-Iwge Classification	Sonographic Characteristics
–	CL	Unilocular anechoic cyst with no wall
Type 1	CL1	Unilocular anechoic cyst with wall and mobile internal echogenicity
Type 2	CE 2	Multivesicular and multiseptated cyst with multiple daughter cysts
Type 3	CE 3	Detached membrane seen within cyst (water-lily sign)
Type 4	CE 4	Heterogeneous (hypo or hyperechoic) cyst with no daughter vesicles
Type 5	CE 5	Cyst having partial or complete wall calcification

Our case presented with right upper quadrant swelling associated with dull aching pain and jaundice during third trimester of pregnancy. An ultrasound was performed which showed a large cystic lesion in liver with daughter cysts containing echogenic debris and crumpled membranes. The cyst was reaching up to sub capsular location s/o hydatid cyst Ghabri type 2. Due to contraindication of CT scan in pregnancy MRI was done which was suggestive of large cystic lesion in liver with multiple daughter cysts, few showing wall calcifications. These features on ultrasonography and MRI confirmed the diagnosis of hydatid cyst of liver.

Treatment of hydatid cyst during pregnancy is difficult due to unavailability of standard or ideal treatment protocol. The treatment options available are medical therapy, percutaneous aspiration by PAIR technique (puncture, aspiration, injection and reaspiration) and conservative surgery. The choice of treatment depends on site, size and type of cyst, symptoms and expected complications.

Medical therapy consists of treatment with anti-helminthic benzimidazolic drugs like albendazole and mebendazole. They are category C drugs i.e.; in animal models these drugs are shown to be embryotoxic and teratogenic. Their use in first trimester is contraindicated due to teratogenic effects leading to facial and limbs abnormalities (Choi et al., 2017). Albendazole has better systemic absorption and penetration into the hydatid cyst. The response of treatment depends on the thickness of the cyst wall and associated calcification. The dose of albendazole is 10-20 mg/kg of body weight in two divided doses with total dose not exceeding 800mg. This course is given for 28 days and repeated after 14 days rest, a total of 3 such course are given (Noori et al., 2020).

Malhotra et al., (2007) in his report on 22 weeks pregnant female having hydatid cyst in left lobe of liver treated her with albendazole till delivery. She had a full-term normal delivery followed by partial pericystectomy 2 months post-partum. Vliet et al., (1995) treated pregnant women with hydatid cyst in late second trimester and third trimester with albendazole therapy and corticosteroids cover during labour.

PAIR is conducted under local anaesthesia under ultrasound or CT guidance. It involves aspiration of cyst under strict aseptic technique using cannula, followed by scolicidal agent (mainly hypertonic saline) injection for 15-20 mins and then reaspiration of cystic contents. This is repeated till the aspirated content is clear. The cyst residua are filled with isotonic normal saline (Noori et al., 2020). PAIR can be used as an effective treatment in second trimester. In third trimester PAIR becomes difficult to perform due to compression effect of enlarging uterus, it is also associated with risk of anaphylactic shock and recurrence of cysts during pregnancy. Moreover, PAIR cannot be done in superficial or pedunculated cysts, small volume cysts and cysts with multiple daughter cysts (Ünalp et al., 2008).

The surgical procedure for hydatid cyst during pregnancy should be conservative like evacuation and de-roofing of cyst or radical approach such as peri cystectomy. Rarely hepatic resection surgery is performed. Timing and indication for surgery should be individualised. WHO does not recommend for hydatid cyst operation during pregnancy. Second trimester is the safest time for performing surgery due to lower risk of abortion and better visualisation of operative field due to smaller size of gravid uterus (Noori et al., 2020). Anaesthesia exposure during the first trimester is associated with higher risk of spontaneous abortion and low birth weight; therefore, any operative procedure should be avoided during the first trimester (Allaert et al., 2007). Surgical treatment may be postponed for 3 months if diagnosis is made in third trimester unless there is some complication like infection or rupture (Manterola et al., 2004).

Reviewing the findings in various studies a decision of giving the patient medical therapy with albendazole was taken and pregnancy was terminated by elective caesarean section after corticosteroids coverage for foetal lung maturity. Post LSCS after 3 months a CT scan was done and liver parenchymal conserving peri cystectomy was performed by the surgery department (Figure 6a, 6b, 6c). The histopathological confirmation of the cyst was done (Figure 7a, 7b, 7c).

Hepatic hydatid cyst is associated various complications like due to pressure generated on adjacent structures and it mostly depends on size and anatomical location of cyst. Obstructive jaundice, cholangitis, pancreatitis are various complications seen in the pancreato-biliary tract due to compressing effect of hydatid cyst. Raised direct bilirubin and icterus points towards obstructive jaundice in our case due to compressing effect of hydatid cyst on the extra hepatic biliary ducts. There was clinical improvement and decrease in bilirubin levels following peri cystectomy in post-partum period due to release of pressure on biliary tract from by the hydatid cyst.

4. CONCLUSION

Human hydatidosis during pregnancy is rare. These pose a serious risk to the pregnant female and the foetus. The management of hydatid cyst is challenging, problematic and not straightforward. Each case needs to be managed on an individual level keeping multiple considerations in mind. Timely diagnosis and appropriate treatment of hydatidosis during pregnancy can lead to a good fetomaternal outcome.

Funding

This study has not received any external funding.

Conflict of interest

The authors declare that there is no conflict of interests.

Data and materials availability

All data sets collected during this study are available upon reasonable request from the corresponding author.

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