

Case Report

Hydatid Disease of Liver during Pregnancy: Report of Two Cases and Review of Literature

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Abstract

Hydatid disease arises from the parasitic infection of *Echinococcus granulosus* and the disease is located most common in the liver. The diagnosis of liver hydatid cysts is usually easy but management of the disease in pregnancy poses some problems. In this article, we report two cases of liver hydatid disease during pregnancy.

Keywords: Echinococcosis; Hydatid Disease; Management; Pregnancy

Introduction

Echinococcosis or hydatid disease is parasitary infection disease and it is caused by the larval form of *Echinococcus Granulosus*. It's primarily a disease of sheep and cattle. Human beings are accidental hosts. Cystic lesions are constituted by parasite, most commonly in the liver and also in other organs. The disease is rare during pregnancy. Main treatment is surgical and medical treatment is necessary in selected cases. So treatment should be individualized for each patient. The diagnosis of hydatid disease of liver is not difficult but management of pregnant women poses some problems. Recurrence of hydatid disease seems more common during pregnancy [1]. In this article, we reported two cases of hepatic hydatid disease in pregnancy; one of them was diagnosed with elevated levels of hepatic transaminases at term and the other was diagnosed with acute abdomen in the 14th week of pregnancy.

Case Report 1

A 36-year-old woman, gravida 4, para 2 was referred to our clinic due to elevated hepatic transaminase levels at the thirty-eighth week of pregnancy. She had no symptoms and current pregnancy has continued uncomplicated until that time. There was no prominent feature in the patient's history. The patient was haemodynamically stable, afebrile and non-icteric at physical examination. In laboratory findings, ALT: 65U/l and AST: 89U/l were detected. The other laboratory investigations were within normal range except for a mild degree of anemia. Blood pressure was stable and proteinuria was not detected in urine analysis. We did not focus on obstetric cholestasis due to lack of pruritus. Obstetric ultrasonography confirmed 38 weeks pregnancy. Hepatobiliar ultrasonography revealed a 58 mm diameter cystic formation with calcified wall in the left lobe of the liver with characteristic appearance for type 5 liver hydatid cyst (Figure 1). Doppler examination revealed no

blood supply in the mass (Figure 2). An indirect haemagglutination test confirmed the diagnosis of a hydatid disease. The patient settled with conservative management and followed in our clinic for one week. Hepatic transaminases persisted at same levels. Consultation from the department of General Surgery was asked for the necessity of removing the mass intra-operatively during the cesarean delivery, but watchful waiting was proposed due to the nature and calcified appearance of the mass. One week later at the 39th week of gestation a 3650 gr, 52 cm, healthy, male infant baby was delivered by cesarean section due to previous cesarean section history. A calcified, solid mass was palpated in the left lobe of liver during the operation. In the postpartum period transaminases persisted at same levels.

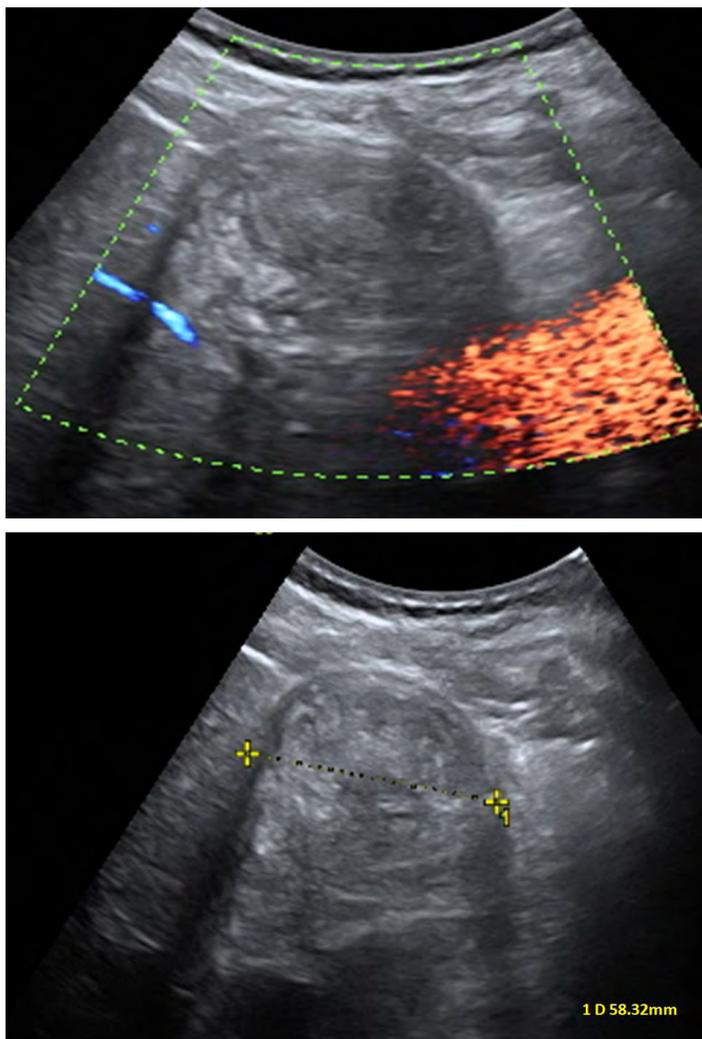


Figure 1, 2: Type 5 liver hydatid cyst in sonography.

Case Report 2

A 30-year-old woman, gravida 3, para 2 was admitted to our clinic with acute abdomen and wide urticarial rashes widespread throughout body. Rashes had arisen suddenly along

with abdominal pain. In obstetric sonography 13weeks non-complicated pregnancy was confirmed. There was no history of trauma. Patient told that abdominal pain was started about one hour ago suddenly at umbilicus and then spreaded to all abdomen. Rashes appeared suddenly throughout all body consecutively to abdominal pain. At physical examination, the patient was moderate in general view, consciousness was clear and she was cooperative. Wide rashes were seen throughout all of her body, especially condensed on chest and upper extremities. Abdominal examination revealed upper abdominal distension with rebound tenderness. Intestinal activity was reduced. Patient was consulted to general surgery clinic for acute abdomen signs. After the evaluation by general surgery clinic, abdomen ultrasonography revealed a 75×50mm cystic formation at the right lobe of the liver that was compatible with cyst hydatid and intraperitoneal free fluid was detected at perihepatic and paracolic areas. Anaphylactic reaction was treated with parenteral methyl-prednisolone and feniramine combination. Then patient underwent laparotomy. Ruptured cyst hydatid and cyst originated free fluid in the peritoneal cavity confirmed at surgery. Cystectomy and drainage was performed. After surgery albendazole 400mg/day therapy was started and decided to continue for minimum three months. Patient is informed about the necessity and risks of albendazole therapy during pregnancy. Patient is now at the 26th week of pregnancy and continues albendazole therapy. Any malformation is not detected on sonography.

Discussion

Hydatid cyst disease is a worldwide parasitic disease that can cause severe morbidity. Hydatid disease is very rare during pregnancy and its incidence is about 1/20,000–1/30,000 [2]. Torsion or rupture of cysts, pelvic inflammation and anaphylaxis are among the complications of the disease. But only less than 5% of the cases are complicated during pregnancy. The disease does not adversely affect pregnancy frequently [3]. Diagnosis can be made by serology and radiological imaging. Ultrasound can be used to avoid radiation exposure in pregnant patients. Usually these methods are adequate for the diagnosis. Also magnetic resonance imaging (MRI) can be used for diagnosis during pregnancy. However MRI is rarely needed for hydatid disease of the liver, while it can be used in non-liver hydatid disease especially musculoskeletal disease [4]. Serological tests for the diagnosis are ELISA, indirect haemagglutination test, and complement fixation test. The decrease in cell mediated immunity in pregnancy can cause rapid progression of the disease and rupture of the cyst may be seen in pregnancy [5, 6]. Anaphylactic reaction may be eventuated with the rupture of cyst. Recurrence of hydatid disease during pregnancy is reported in many cases, in patients with a history of hydatid disease [7]. The disease is endemic in Turkey and humans are most frequently infected by dogs [8]. Hydatid cyst disease causes such several symptoms and signs or it may be

asymptomatic and may be detected randomly. Our first patient was diagnosed randomly with mild elevated transaminase levels and the diagnosis was done by sonography. The second case is diagnosed dramatically with acute abdomen signs and rashes on the body. Ultrasonography confirmed cystic hydatid disease of the liver. Then the patient was taken to the surgery emergently where cystectomy and drainage were performed.

The ultrasonographic definition of hydatid disease of the liver is classified to five types by Gharbi in 1981. Type 1: Pure fluid collection, Type 2: Pure fluid collection with separated wall, Type 3: Septated fluid collection, Type 4: Heterogeneous mass with rough echo, Type 5: Thick and calcified wall. Type 1-2 are active lesions, type 3 is transient and type 4-5 are inactive lesions [9].

Although surgery is the mainstay treatment of cystic hydatid disease of the liver, medical treatment should be the first choice for patients who have too many cysts, who cannot tolerate the surgery and who do not consent to the surgery [10]. Medical treatment is primarily albendazole therapy. We used 400mg/day albendazole therapy for the second patient after surgery and continued albendazole for three months after surgery due to rupture of the cyst. Albendazole is classified in category 'C' (animal studies have shown an adverse effect on the fetus and there are no adequate and well controlled studies in humans) during pregnancy and it is not preferred in the first trimester due to the risk of teratogenicity including limb defects and facial abnormalities [11]. We followed the first patient conservatively avoiding surgery or medical treatment because it was type 5 liver hydatid cyst and had a calcified wall. Other defined procedures in the treatment include PAIR (puncture, aspiration, injection and re-aspiration) technique. This technique is an ultrasound guided technique that consists of puncture and evacuation of the contents of the hydatid cyst, injection of a scholoidal agent such as 95% ethanol and reaspiration of the contents of the cyst [12]. Ustunsoz et al reported six patients successfully treated by PAIR technique during pregnancy. They used hypertonic saline solution as cytotoxic agent and they did not use albendazole prophylaxis [13].

There is no clear suggestion regarding the intrapartum management of patients affected by hydatid disease because of the rare nature of this condition. Both successful vaginal and operative deliveries have been reported in literature. But these lesions may involve the female pelvis that can cause complicated delivery and may result in obstructive labor. So intrapartum management should be individualized [14].

Transplacental transmission of the disease to the fetus or neonate seems impossible. Manterola *et al.* showed that by placental histology and serological tests on the neonates of affected mothers in Chile [15].

In summary, hydatid disease may be encountered with such

symptoms and signs in pregnancy. It should be kept in mind especially in endemic countries and management should be individualized for every patient.

Conflicts of Interests: There is no declaration of conflicts of interest.

References

1. Kain KC, Keystone JS. Recurrent hydatid disease during pregnancy. *Am J Obstet Gynecol.* 1988, 159(5): 1216–1217.
2. Rahman MS, Rahman J, Lysikiewicz A. Obstetric and gynaecological presentations of hydatid disease. *Br J Obstet Gynaecol.* 1982, 89(8): 665–670.
3. Jackisch C, Schwenkhagen A, Louwen F, Raber G, Oehme A et al. Echinococcosis in pregnancy. A rare differential diagnosis in cystic intra-abdominal tumors in pregnancy. *Zentralbl Gynaekol.* 1993, 115(6): 263–272.
4. Durakbasa MO, Kose O, Islam NC, Kilicoglu G. A primary hydatid cyst of the gracilis: a case report. *J Orthop Surg (Hong Kong).* 2007, 15(1): 118-120.
5. Dede S, Dede H, Gokcu M, Gelisen O. Recurrent echinococcal disease of liver in pregnancy: A case report. *Gynecol Obstet Reprod Med* 2005; 11:72-73.
6. WHO Informal Working Group. International classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings. *Acta Trop.* 2003, 85(2): 253–261.
7. N Malhotra, C Chanana, S Kumar. Hydatid Disease Of The Liver During Pregnancy: A Case Report And Review Of Literature. *The Int J Gyn Obs.* 2006,7(1).
8. Sahin E, Nayki U, Sadik S, Oztekin O, Nayki C et al. Abdominal and pelvic hydatid disease during pregnancy. *Arch Gynecol Obstet.* 2005, 273(1): 58-59.
9. Gharbi HA, Hassine W, Brauner MW, Dupuch K. Ultrasound examination of hydatid liver. *Radiology.* 1981, 139(2): 459-463.
10. Zeybek B, Ergenoglu AM, Yeniel AO, Ulukus M. Hepatic hydatid disease during pregnancy. *Ege Journal of Medicine.* 2010, 49(3): 197-199.
11. Moussa AM, Mehrez MG, Muhtaseb SA, AL-Mudala DS, Kamel SM. Disseminated pelvic hydatidosis presenting as ovarian carcinomatosis: successful post-operative treatment with mebendazole. *Int J Gynaecol Obstet.* 1987, 25(6): 473-478.
12. Bilgic S, Kose O, Sehirlioglu A, Safaz I, Ozkan H. Primary

paraspinal hydatid cyst treated with puncture, aspiration, injection, and reaspiration (PAIR) technique: A case report. *Eur Spine J.* 2009, 18(2): 165- 167.

13. Ustunsoz B, Ugurel MS, Uzar AI, Duru NK. Percutaneous treatment of hepatic hydatid cyst in pregnancy: long-term results. *Arch Gynecol Obstet.* 2008, 277(6): 547-550.

14. Dede S, Dede H, Caliskan E, Demir B. Recurrent pelvic hydatid cyst obstructing labour, with a concomitant hepatic primary. *Acase report. J Reprod Med.* 2002, 47(2): 164-166.

15. Manterola C, Espinoza R, Munoz S, Vial M, Bustos L et al. Abdominal echinococcosis during pregnancy: clinical aspects and management of a series of cases in Chile. *Trop Doct.* 2004, 34(3): 171-173.