Hepatic hydatid disease during pregnancy
Gebelikte karaciğer kist hidatığı

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Summary
Hydatid cysts most commonly arise from infection from Echinococcus granulosus and their most common site is the liver. The diagnosis of liver hydatid cysts is not difficult but management modalities of pregnant women pose some problems. Although both medical and surgical treatments are available, there is no consensus. In this article, we report a hepatic hydatid disease in a pregnant woman who was managed with albendazole therapy.

Key words: Hydatid disease, pregnancy.

Özet

Anahtar kelimeler: Kist hidatik, gebelik.

Introduction
Hydatid disease or Echinococcosis is caused by Echinococcus granulosus or Echinococcus multilocularis. The incidence in pregnancy ranges from 1 in 20,000 to 1 in 30,000 (1). A hydatid cyst is mainly encountered in countries where cattle and sheep raising are important, such as Argentina, Australia, Chile, South America, India and Turkey (2). Infected dogs are frequently responsible for human contamination (3). Hydatid cysts may remain asymptomatic for many years and may be found incidentally on imaging. These cysts during pregnancy are very rare and the choice between treatment modalities such as surgery, medical treatment or percutaneous drainage is still controversial.

Case Report
A 27- year- old gravida 2, para 1, obstetrical history of one previous cesarean section, 30-week- pregnant woman was admitted to our clinic with epigastric and right upper quadrant pain. On examination the patient was hemodynamically stable, afebrile, with tenderness at the right upper quadrant. Rebound and musculer defence were not present. Haematological and biochemical results were normal. An ultrasound of the upper abdomen revealed a 14x10 cm cystic mass at sector 7 and a 7x5 cm cystic mass at sector 6 of liver. The rest of the ultrasound examination was normal. A 30- week- fetus with positive fetal cardiac activity was noted in the uterus. An indirect haemagglutination test and ELISA test both confirmed the diagnosis of a hydatid disease. The patient decided on conservative management and the next day pain at the right upper quadrant regressed. General Surgery consultation offered albendazole therapy 400 mg, twice daily.
Surgery was avoided from operation because of the risk of preterm labour and severe anaphylaxis during surgery. The cyst neither progressed nor regressed during the remainder of pregnancy. At 38 weeks gestation she gave birth to a female baby weighing 3030 gr via caesarean section. The placenta did not reveal any abnormality on gross examination and histopathology. The baby’s ELISA and indirect hemagglutination tests were same as the mother’s and was considered passive immunization. Since 10 days after delivery, she has been still in follow up with albendazole therapy. General surgery will offer surgery after the postpartum period which is six weeks after delivery.

**Discussion**

There are two species of Echinococcus. Echinococcus granulosus which causes cystic Echinococcosis and Echinococcus multilocularis which causes alveolar Echinococcosis. The ova are ingested by the human via a faeco-oral route from infected canines and hatch to form larvae in the small intestine. After penetration of the intestinal wall and blood vessels, hydatid cysts can occur anywhere in the body such as orbit (4), left ventricle of the heart (5) and ovaries (6) although the most common presentation site is the liver. The decrease in cellular immunity during gestation may explain the rapid progression of the disease (7). In the present case, the patient did not indicate any risk factors.

As for pregnant woman, complications such as abdominal pain, dystosis, and uterine rupture which vary according to the involvement site and dimensions of the cyst, may be seen. Jasper (8) reported one case of hydatid disease which resulted in dystocia during obstetric labour. The patient had a pelvic hydatid disease diagnosed at the time of caesarean section. Moreover it has been reported that patient deaths may occur due to anaphylaxis, which results from rupture of cysts during labor (9).

Diagnosis of hydatid disease is made by radiology and serological testing which confirms the diagnosis. Ultrasound can be used to avoid radiation exposure in pregnant patients. The most commonly used serological tests are ELISA, indirect haemagglutination test (IHA), and complement fixation test (CF). The CF test returns to negative after successful treatment and is important in postoperative follow up. The most accurate serological test is immunoelctrophoresis, as it is positive only with echinococcus granulosus and echinococcus alveolaris and is based on the interaction of the serum from the hydatid-infected patient with the antigen compared to a control.

The baby’s follow-up is challenging as the anticores can be detected for 15 months postpartum. Avidity index is helpful in this period as high levels support the maternal passive immunization during gestation. Low levels may suggest an acute infection, although there have been no reported cases of transplacental transmission in the literature. For accurate diagnosis, ELISA and IHA can be repeated at 15-18th months.

Management includes three different modalities: 1. Medical treatment 2. PAIR (puncture, aspiration, injection and re-aspiration) technique 3. Surgery. Although surgery is the mainstay treatment of cystic hydatid disease of the liver, medical treatment can be the first choice in patients who have too many cysts, who can not tolerate the surgical treatment and who do not consent to the surgery. Medical treatment consists of Albendazole therapy. According to the United States Food and Drug Administration (FDA), 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) and it is not preferred in the first trimester due to the risk of teratogenicity including limb defects and facial abnormalities. The optimum dosage is 15 mg/kg divided into two doses daily (max 800 mg daily) in pregnancy. The Albendazol regimen consists of 4 weeks of medication and 2 weeks of breaks and this scheme is repeated three times. During the lactation period, it is classified in the ‘unknown’ category although there is no report of an adverse effect in the literature. Response to Albendazole depends on thickness of the cyst wall and the absence of calcifications. Cysts recurring after surgery or cysts that do not demonstrate regression after medical treatment may be treated with the PAIR technique. This is an ultrasound guided technique consisting of puncture and evacuation of the contents of the hydatid cyst, injection of a scholosidal agent such as 95% ethanol, and reaspiration of the contents of the cyst (10). Although the procedure is highly effective in intact univesicular and multivesicular cysts, removal of all the viable cyst elements can be very difficult and potential disease could be left behind in the cyst cavity which could become infected. In the literature, percutaneous treatment often requires albendazole coverage to counteract possible intraabdominal spillage.

Different surgical approaches such as simple cyst evacuation or partial pericystectomy, or radical surgeries such as liver resection or cystopericystectomy have been reported. Golaszewski (11) reported a case of a
primigravida who was diagnosed to have a large, 20 cm hydatid cyst of liver at 14 weeks of gestation. The patient successfully underwent subtotal cystectomy under anti-helmintic cover at 19 weeks of gestation. Surgeons from endemic areas tend to favour conservative management whereas those outside the endemic areas tend to favour radical surgery. The optimal timing of surgery in a pregnant patient is the second trimester because there is a lower risk of spontaneous abortion and the uterus is not in the way of the operative field. Surgical management has been associated with a recurrence rate of approximately 10% (12). Preoperative and postoperative Albendazole use prevents this relatively high recurrence rate. As our patient was 30 weeks pregnant, general surgeons avoid surgery. As a result the treatment of hydatid disease is difficult during gestation and neither surgical nor medical treatments are completely safe.

Kaynaklar