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Retrospective Evaluation of Hydatid Cyst Cases During Pregnancy

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Aim: Zoonotic parasite infections affect many pregnant people around the world. Hydatid cystic disease is also a zoonotic disease caused by *Echinococcus* sp. This study aims to present the maternal-fetal results and clinical treatment of pregnant women diagnosed with liver hydatid cyst (CH). This zoonotic disease is discussed again in the light of current literature. **Materials and Methods**: Pregnant women with hydatid cyst monitored in a tertiary health center between 2018 and 2020 were evaluated. Seven cases were included in this study. We retrospectively collected and analyzed clinical data, which did not interfere with medical treatment. **Results**: Albendazole was started as medical therapy in six patients, and percutaneous drainage was applied to one patient. Three of our six patients who started medical treatment had to undergo surgery due to maternal complications that developed despite medical treatment. Two of our patients were delivered with a cesarean section due to the obstetric indications. **Discussion**: Hydatid cysts are most commonly caused by *Echinococcus granulosus* infection and most common in the liver. The diagnosis of liver hydatid cysts is not difficult, but pregnant women's treatment methods have some problems. Although both medical and surgical treatments are available, there is no consensus. We would also like to underscore that echinococcal disease of the liver should be kept in mind in the differential diagnosis of abdominal pain, jaundice, and/or fever, especially in endemic regions. We think that when we increase awareness about this disease, we can improve fetal and maternal outcomes by making an early diagnosis and management.

INTRODUCTION

Zoonotic parasite infections affect many pregnant women around the world. Parasite infections have maternal, fetal, and placental effects. These infections are associated with adverse outcomes such as anemia and malnutrition. Parasite and tropical infections generally affect pregnant women more than non-pregnant women. Fetal and placental effects in the first trimester compared to the next periods of pregnancy is more serious. Immune system changes during pregnancy increase susceptibility to parasitic infections [1,2]. Hydatid cystic disease is also a zoonotic disease caused by the *Echinococcus* sp. tapeworm (Figure 1) [3]. The definitive hosts are carnivores, including dogs, foxes, and wolves, and the intermediate hosts are herbivores, including sheep, cattle, and deer. Humans act as accidental hosts [4]. Hydatid cysts are found more commonly in sheep-rearing areas globally, namely

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Abbreviations: CH, hydatid cyst; ELISA, enzyme-linked immunosorbent assay; IHA, indirect hemagglutination test; CF, complement fixation test.

Keywords: Hydatid cysts, Pregnancy, *Echinococcus granulosus*, Maternal-Fetal outcomes

Figure 1. Microscopic view of *Echinococcus granulosus*.

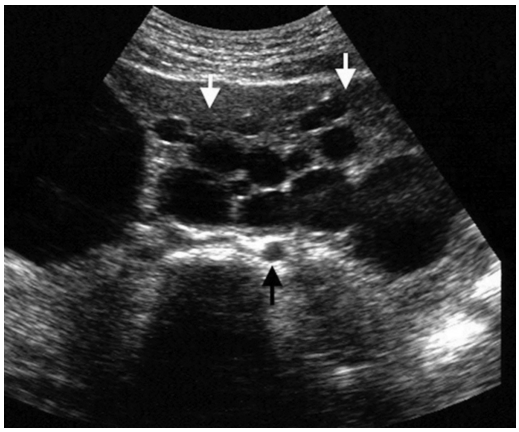

Figure 2. Radiologic Features of Hydatid Disease.

Australia, New Zealand, Argentina, Chile, India, and the Mediterranean. Hydatid cysts may remain asymptomatic for many years and maybe discovered incidentally on imaging. These cysts can be found in any organ, but lung and liver hydatids are the most common (Figure 2) [5,6]. Hydatid cysts during pregnancy are rare, and also both surgical and medical modalities have been tried.

This current study aims to present the maternal-fetal outcomes and clinical treatment of pregnant women diagnosed with hydatid cyst (CH) of the liver. This zoonotic disease is discussed again in the light of current literature.

MATERIALS AND METHODS

Pregnant women with hydatid cysts that were monitored in a tertiary health center between 2018 and 2020 were evaluated. Seven cases were included in this study. We retrospectively collected and analyzed clinical data, which did not interfere with medical treatment. The local ethic committee granted its approval for the conduct, protocol, and procedures of this research article. The follow-ups of the patients were performed by obstetrics and gastroenterology departments. Diagnostic tests were performed in the presence of clinical signs. The diagnosis of hydatid disease is made by ultrasonography and serological testing, which confirms the diagnosis. Enzyme-linked immunosorbent assay (ELISA), indirect hemagglutination test (IHA), and complement fixation test (CF) were used as serological tests. Surgery, medication, or cyst drainage was performed for treatment. Concurrently, the biophysical evaluation of fetuses and follow-ups were performed with obstetric ultrasonography.

STATISTICAL ANALYSIS

Data analysis was performed using IBM SPSS Statistics 20.0 software (IBM Corp. Released 2011. IBM SPSS Statistics for Windows, Version 20.0. Armonk, NY: IBM Corp.). Our categorical data included included gestation, delivery week, birth weight, and APGAR score and were evaluated using descriptive statistics for analyzing. Descriptive statistics for these continuous variables were presented as mean ± standard deviation (SD).

RESULTS

Cystic hydatid was detected incidentally in only one of our seven patients. The main complaint in our other six patients was abdominal pain (85%). Two of our patients had jaundice at the time of admission to the hospital. All of our patients were in the 2nd or 3rd trimester at the time of diagnosis. Four of our patients were delivered by C-section (66%). The average birth weight was 2591.66 ± 715.83 gr. The mean of 1st minute APGAR score was 7.33 ± 1.03. The mean birth delivery time was 35 ± 3.57 week. The mean of the 5th minute APGAR score was 8.83 ± 0.98. One patient, whose diagnosis was at the earliest gestational week (14th week), also had dyspepsia complaints. Albendazole was started as a medical treatment in six patients, and percutaneous drainage was applied to one patient. Three of our six patients who started medical treatment had to undergo surgery due to maternal complications that developed despite medical treatment. Two of our patients were delivered by cesarean due to obstetric indications. One patient had sepsis and fever unresponsive to medical treatment in the post-diagnosis observation. Since surgical treatment was decided, cesarean delivery was performed in the same session. In our patient whose dyspeptic complaints were accompanied by jaundice and abdominal pain, it was found that intraabdominal abscess developed in the ultrasonographic examination. Abscess drainage was performed surgically. In the postoperative controls, the patient was observed to have had a miscarriage. In our other patient who had to undergo surgery, since cystic hydatid rupture was detected at the time of diagnosis, she was taken to the

Table 1. Clinical Features of our Cases.

Case Number	Pregnancy Week in the Diagnosis	Initial Symptoms	Treatment	Delivery Week	Delivery Type	Maternal Complication	Fetal Complication	Birth Weight	Apgar 1st Minute	Apgar 5th Minute
1	28	Abdominal pain	Albendazol	39 week	C/S	No	No	3400 gr	8	10
2	30	Jaundice+ Abdominal pain	Surgery+ Albendazol	30 week	C/S	Fever and Sepsis	Prematurity	1800 gr	6	8
3	16	Incidental	Albendazol	37 week	C/S	No	No	3100 gr	7	9
4	30	Abdominal pain	Albendazol	38 week	Vaginal	No	No	3200 gr	9	10
5	14	Dyspepsia+ Abdominal pain+ Jaundice	Surgery+ Albendazol	Abortus in 16 week	Vaginal	Abscess	-	-	-	-
6	32	Abdominal pain + mass	Surgery+ Albendazol	32 week	C/S	Cyst Rupture	Prematurity	1950 gr	7	8
7	22	Abdominal pain	Drainage	34 week	Vaginal	No	Prematurity	2100 gr	7	8

Table 2. Treatment modalities of the *Echinococcus* species.

Treatment	Indications
Surgery	• Complicated cysts (eg, rupture cyst, cysts compressing vital structures, cysts with biliary fistulae, cysts with hemorrhage or secondary infection) • Cyst diameter >10 cm • Superficial cyst at risk of rupture due to trauma • Extrahepatic disease (lung, brain, kidney, bone, or other site)
Percutaneous management	• PAIR technique → destroy the germinal layer with scolicidal agents • Evacuating the entire cyst → cysts that are difficult to drain, relapse after PAIR
Drug therapy	• Albendazole is the primary antiparasitic agent • Definitive management in selected cases • Useful adjunctive therapy to surgery and percutaneous treatment

Table 3. Clinical manifestations the *Echinococcus* species.

Liver Involvement	Lung Involvement
fever	fever
right upper quadrant pain	cough
nausea	chest pain
vomiting	dyspnea
dyspepsia	hemoptysis
obstructive jaundice	malaise
biliary colic	nausea
cholangitis	vomiting
pancreatitis	thoracic deformations
cholestasis	pneumothorax
portal hypertension	pleural effusion
Budd-Chiari syndrome	empyema
liver abscesses	pulmonary abscess
sepsis	sepsis

emergency cesarean section, and the hydatid cyst capsule was removed in the same session. When we evaluated the fetal results, prematurity was observed in babies of pregnant women who underwent a cesarean section due to maternal complications at 30 and 32 weeks of gestation. In addition, our patient, upon whom we performed percutaneous drainage, performed preterm delivery at the 34th week (Table 1).

DISCUSSION

We have minimal data about the treatment, follow-up, and complications of hydatid cyst in pregnancy. A small number of cases with liver CH during pregnancy have been reported in the literature [7].

There are three treatment modalities (Table 2). These are systemic medication, surgery, and percutaneous drainage. However, surgical treatment is the first option among these. Medical treatment is applied in uncomplicated CH, in the presence of too many cysts, in patients who cannot tolerate surgical treatment, and in patients who do not accept the operation. The drug of choice in medical treatment is albendazole [8]. Furthermore, where parasite endemicity is 20%, the World Health Organization (WHO) recommends preventive chemotherapy with single-dose anthelminthic drugs: albendazole or mebendazole. There are concerns about the usage of a wide range of anthelminthic drugs by women of reproductive age, including pregnant women, after the first trimester, particularly in the first few weeks when the pregnancy may not yet be confirmed, according to WHO [9].

CH may grow during pregnancy due to suppression of cellular immunity and steroids secreted from the placenta. The cyst may grow and rupture. The patient's general condition deteriorates, shortness of breath, and chest pain increase, with cyst rupture. Sometimes deaths may occur due to anaphylactic shock and bleeding [10,11]. The most frequent symptom was abdominal pain in our pregnant women, and prematurity was the most frequent fetal complication due to preterm birth. General clinical symptoms have been listed in our table (Table 3).

Considering the publications on pregnancy and hydatid cysts in the literature, there is a 16-week pregnant woman with a diagnosis of liver CH who was treated percutaneously and completed her pregnancy without any complications [12]. Besides, Sahin et al. reported another 19-week-old patient with liver and pelvic CH disease, who was treated with laparotomy without complications during her pregnancy [13]. There are few studies about this topic [14]. Our study is one of the largest series with seven cases in the literature.

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 pregnant women's management modalities pose some problems. Although both medical and surgical treatments are available, there is no consensus. During pregnancy, these cysts are very rare, and the choice between treatment modalities such as surgery, medical treatment, or percutaneous drainage is still controversial. If parasitic infections are detected early during pregnancy and treated appropriately, the results improve significantly. It is essential to focus on parasitic infections during pregnancy and pay attention to those who travel. CH develops slowly in humans and progresses without symptoms in most cases. Considering that clinical symptoms

can take a long time to emerge, the importance of protecting children from contact with infected animals and teaching cleanliness awareness at an early age becomes evident. Raising the public's awareness about zoonoses and preventing the transmission routes between sheep, dogs, and humans as much as possible is considered to be the most important precaution to be taken against the disease.

In this paper, we want to emphasize that as a reminder that echinococcal disease of the liver must be included in the differential diagnosis of abdominal pain, jaundice, and/or fever occurring in pregnancy, especially in endemic areas. When we increase awareness about this disease, we think that we can improve fetal and maternal outcomes by making an early diagnosis.

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