

## Echinococcosis of the Spleen during Pregnancy

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Echinococcosis, also termed hydatid disease, is an endemic disease on the Asian continent. Israel is one of the endemic regions. Caused by the tapeworm *Taenia echinococcus*, in our region the common variant is *E. granulosus*, which produces cysts [1]. The majority of these cysts (50–80%) involve the liver, while less commonly affected organs are the lungs, kidneys and spleen. Isolated hydatid disease of the spleen is rare, even in endemic areas, constituting 0.5–4% of all cases of echinococcosis [2].

Very few cases of echinococcosis in pregnancy have been reported and most of them involved the liver or lungs [3]. The patient described here is, to the best of our knowledge, the first reported case of echinococcal cyst of the spleen during pregnancy.

### Patient Description

A 27 year old Arab woman living in a rural area was admitted to our surgery department because of a large asymptomatic upper abdominal mass that had been noted 6 months earlier and had enlarged significantly during the last month. The patient was pregnant (5 weeks gestational age). Her obstetric history revealed one normal vaginal delivery and one spontaneous abortion due to unknown cause. Her medical history was unremarkable.

Physical examination was normal, except for a large, rounded, immobile painless mass palpated in the left upper abdominal quadrant and extending to the epigastrium. Laboratory analyses disclosed a positive beta-human chorionic gonadotropin test, normocytic normochromic anemia (hemoglobin 10.1 g/

dl), leukocyte count 5,110/mm<sup>3</sup> and platelet count 224,000/mm<sup>3</sup>. Liver and renal function tests were within normal limits.

Abdominal sonography confirmed the cystic nature of the palpated mass and its intimate anatomic relationship to the spleen, and showed medial compression of the left lobe of the liver and downward displacement of the left kidney. The cystic wall was thin without evidence of daughter cysts within [Figure]. Otherwise, the abdominal examination was normal and the uterus was consistent with pregnancy.

No further tests were deemed advisable or necessary due to the early pregnancy, and midline laparotomy was performed. The intraoperative findings were compatible with the preoperative ultrasound examination. The huge cyst involved the splenic hilus and upper

pole, extended medially compressing the liver, and was adherent to the left diaphragm. During open drainage of the cyst fluid an anaphylactic reaction occurred, probably induced by some spillage, and blood pressure had to be stabilized with intravenous hydrocortisone. At this stage the diagnosis of hydatid cyst was obvious. The germinal layer was excised and the cyst and abdominal cavities were thoroughly irrigated with hypertonic saline. Following unroofing of the cyst omentoplasty was performed.

Postoperative gynecological examination and fetal monitoring were normal. The early convalescence was complicated by generalized urticaria, which necessitated intravenous hydrocortisone for 2 days and upper extremity thrombophlebitis that resolved under local alcohol compresses and systemic anti-



Huge splenic cyst as demonstrated by ultrasonography.

biotic treatment. Pathological examination of the cyst wall confirmed hydatid disease.

It was decided that albendazole treatment be administered and that the pregnancy be terminated. This decision permitted us to perform an abdominal computed tomography scan, which revealed a deep small echinococcal cyst in the right lobe of the liver, further strengthening the indication for the albendazole course. Left subphrenic collection was also diagnosed and percutaneously drained under CT guidance. When the thrombophlebitis and subdiaphragmatic collection subsided, the pregnancy was terminated and further convalescence was uneventful.

## Comment

Throughout the world echinococcal disease is the most frequent cause of liver cysts [1]. It is mainly encountered in sheep-raising areas where dogs have access to infected offal and are infested by eating the viscera of sheep. Humans, serving as an intermediate host, are infested by ingestion of food contaminated by eggs excreted by the definitive host [1,3].

There is little knowledge on the natural history, treatment and complications of echinococcal infection during pregnancy, due to limited reported experience [3]. This disease may be asymptomatic throughout the course of pregnancy, while the first presenting sign may be delay in the delivery process caused by the mass effect of the cyst. Abdominal sonography and CT scan are the basic means for diagnosis. Recently a new immunological method to verify the diagnosis was described [2]. Most investigators agree that surgery should be the mainstay of treatment in operable cases, while albendazole or mebendazole should be used preoperatively to decrease the cyst size, decrease the number of daughter cysts, and minimize the risk of anaphylactic response to leakage or rupture. A postoperative course of anti-helminthics is indicated to decrease the risk of recurrence.

In pregnancy the use of these anti-

helminthic agents is relatively contraindicated due to the lack of sufficient data on this subject. Thus early surgery is probably the treatment of choice. This approach is also further supported by the potential risk of cyst rupture during pregnancy due to increasing intraabdominal pressure caused by the enlarging uterus. Impaired cellular immunity during pregnancy might also contribute to rapid echinococcal cyst enlargement. The risk of uncontrolled cyst rupture and the resultant severe consequences are even more threatening during delivery. These considerations and the very large size of the cyst in our case precluded an expectant approach with courses of albendazole after the first trimester of pregnancy [3].

Different surgical techniques have been used to treat splenic hydatid disease. Some support the conservative approach such as partial splenectomy, cyst enucleation and unroofing with omentoplasty, while others prefer total splenectomy [1]. Percutaneous drainage of hydatid cysts during pregnancy may also be an option, however spillage of the cyst content with anaphylactic reaction, as was the case in our patient, might be more difficult to contain and control.

In our case the preoperative differential diagnosis did not place echinococcal cyst as the first choice, since isolated splenic location is very rare (the concomitant lesion in the liver was missed by the ultrasound), there were no characteristic sonographic features, and there was no history of echinococcosis. Even if preoperative serological tests had indicated hydatid disease, despite mechanical considerations due to the very large size of the cyst and the uncertainty concerning the safety of using anti-helminthics at such an early stage of pregnancy, our decision to operate would not be affected. Thus, we also considered the possibility of other cystic lesions of the spleen, and because the patient wished to continue the pregnancy we proceeded to laparotomy without CT scan or serological workup. Intraoperative diagnosis of hydatid disease and the postoperative CT scan, which revealed

the small hepatic lesion, dictated the need for adjuvant treatment with albendazole and the pregnancy was terminated.

The epidemiology of echinococcal disease in Israel has changed during the past 50 years. Shifts in population patterns since 1948 have profoundly affected the distribution of hydatid disease; for example, the mass immigration of Jews from North Africa and Asia Minor reversed the prevalence from the non-Jewish community towards this new population. However, during the last three decades the endemicity has again gradually increased in Arab and Druze communities, especially in northern Israel. Currently, some Druze settlements are among the most highly endemic areas for hydatid disease in the world. These prevalence patterns may be related to home slaughter of sheep, differing attitudes to dogs, and the hunting of wild pigs [4,5].

We believe that in endangered regions the most important aspect of containing and preventing this endemic infection is public health education of the population at risk. This education program should emphasize the role of animals in passing the infection and the necessity of treating infected animals.

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