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# Management of Complicated Hydatid Cyst in Pregnancy. A Multicenter Study

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## Abstract

**Background:** The hepatic hydatid cyst is a major public health concern in endemic areas. It presents a therapeutic challenge when it occurs in pregnant women and exposes the mother and the fetus to a high mortality risk in the case of complications. The main complication is communication with the biliary tree, which fortunately is rare.

**Methods:** A multi-center retrospective study was conducted spanning 7 years, from January 2009 to December 2015. In 17 departments of surgery across Tunisia, 24 cases were identified of pregnant women who were treated for complicated hepatic hydatid cyst. The data on their treatment and outcome were retrieved from the medical records.

**Results:** The age range of the 24 patients was 23 to 40 years, median 30 years. The median gestational age was 15 weeks (range 5 to 29 weeks).The patients complained of a variety of symptoms, mainly abdominal pain (87.5%), fever (50%), jaundice (50%) and vomiting (21%). The laboratory examination showed leukocytosis (54.2%), cholestasis (41.7%), elevated liver enzymes (12%), and positive blood culture (6%). On abdominal ultrasound (U/S), dilatation of the common bile duct was observed in 41% of the cases, dilatation of the intra-hepatic bile ducts in 50%, and hydatid material in the duct in 12% of the cases. Most of the cysts were located in the right lobe of the liver, in the hepatic dome. All of the patients were treated surgically, by one of three types of intervention: Largot intervention (18 cases), internal transfistulary drainage (4 cases), and the Perdromo procedure (2 cases). Postoperative follow-up was complication-free for 20 patients. In the remaining four, complications included: purulent retention (1 case), biliary fistula (2 cases), pneumonia (1 case). There was no maternal death. A tocolytic agent was administered to 16 women and 21 had a live full-term birth. One spontaneous miscarriage and 2 neonatal deaths occurred.

**Conclusions:** Complicated hydatid cyst in pregnancy is a serious condition which can be life-threatening for the mother and the fetus. The treatment is surgical, and patient management requires close collaboration between the surgeon, the anesthesiologist and the obstetrician-gynecologist.


**Key words:** *Hepatic hydatid cyst; liver; pregnancy; Largot; Perdromo; internal transfistulary drainage*

## Introduction

The hepatic hydatid cyst is a disease caused by the parasite *Echinococcus granulosis*, which is endemic in the Tunisian territory. Humans are considered an accidental intermediary host, infected by the consumption of contaminated food. The hydatid cyst can affect any organ, predominantly the liver and the lungs. Hepatic hydatid cyst is rare in pregnant women, but when it occurs it presents a diagnostic and therapeutic challenge, and the complicated forms, with communication between the cyst and the biliary tree, and infection, are life-threatening for both the mother and her fetus.

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## Methods

A multi-center retrospective study was conducted spanning 7 years, from January 2009 to December 2015. In 17 departments of surgery across Tunisia, 24 cases were identified of pregnant women who were treated for complicated hepatic hydatid cyst. The data on their treatment and outcome were retrieved from the medical records.

## Results

The characteristics of the 24 pregnant women with complicated hepatic hydatid cyst are shown in Table 1. Their mean age was 30±1 years, with a range of 23 to 40 years. Most of the women were multiparous (87.5%). The median gestational age at diagnosis of hydatid cyst was 15

**Table 1.** *Characteristics of women with complicated hepatic hydatid cyst in pregnancy (n=24).*

| | |
|---|---|
| Age (years) | |
| Mean | 30±1 |
| Range | 23-40 |
| Gravidity | |
| Parity 1 | 7 (29.2%) |
| Gestation≥2 | 17 (70.8%) |
| Parity | |
| Primiparous | 3 (12.5%) |
| Multiparous | 21 (87.5%) |
| Pregnancy trimester | |
| Trimester 1 | 9 (37.5%) |
| Trimester 2 | 14 (58.3%) |
| Trimester 3 | 1 (4.2%) |

weeks (range 5 to 29 weeks). All the patients were treated surgically at a mean duration of pregnancy of 15.8±1.3 weeks. The procedure was performed on 14 patients in the second or the third trimester of pregnancy.

The presenting symptoms are shown in Table 2. Abdominal pain, fever and jaundice were the most common symptoms.

Leukocytosis was found in 54.2%, cholestasis in 11, and elevated liver enzymes in 5 cases only [10,11]. All patients underwent abdominal ultrasound (U/S), but not computed tomography (CT). The U/S showed dilatation of the common bile duct in 11 cases and dilatation of the intra-hepatic bile ducts in 12 cases. The presence of hydatid material in the bile duct was detected in 5 cases. The cyst was located in the right lobe of the liver, in the hepatic dome, in 11 cases, in the left lobe in 5 cases and in the 5$^{th}$ and 6$^{th}$ segments in 8 cases.

## Treatment

All 24 patients underwent surgery, via right subcostal incision in 22 patients and medial laparotomy in 2 patients.

**Table 2.** *Presenting symptoms in pregnant women with hepatic hydatid cyst (n=24).*

| Symptom | Percentage |
|---|---|
| Abdominal pain | 87.5 % |
| Fever | 50 % |
| Jaundice | 50 % |
| Vomiting | 21 % |

During surgery, the cyst liquid was observed to be purulent in 10 cases, bilious in 10 cases and clear in 2 cases. The common bile duct was dilated in 13 cases. Cholangiography was performed in one patient, showing the common bile duct with heterogeneous content. A Largot intervention was performed in 11 cases, along with omentoplasty in 7, 5 of whom had a small cysto-biliary fistula that was stitched. In 17 patients cholecystectomy was performed.

An internal trans-fistulo-oddian drainage was performed in 4 cases and a Perdromo procedure in 2 of these. Choledocotomy and T tube drainage was performed in 6 cases. A brief description of the surgical procedures is given below.

## Surgical techniques

In all surgical procedures, the patients are placed in the supine position and are operated on by laparotomy. The liver is explored to identify the number and the location of the cysts. The cyst is exposed and isolated from the remainder of operative field, using large abdominal packs soaked in scolocidal agents. The cyst is punctured with a Devé trocar and the color of the hydatid fluid is noted. The puncture opening is secondarily enlarged to ensure complete removal of the cyst and adequate exploration of the residual cavity in order to search for a possible large biliary communication (diameter >5 mm).

- The Perdromo procedure is performed for fistulization of a large cysto-biliary fistula. After the common bile duct has been cleared, a T-tube is placed through a choledechotomy and the fistula is intubated with maximal friction. The tube has a short path inside the cavity, not longer than 2 cm. Another intra-hepatic path is of a least 3 cm long. To obliterate the remaining cavity, a portion of omentum with a good blood supply is sutured into the residual cavity (omentoplasty) and tube drainage is placed. This intervention is indicated for large cysts with a large cystobiliary fistula and a calcified pericystium.
- Unroofing and endocystectomy, or Largot procedure, consists of excising the projecting part of the pericyst. Hemostasis and bilistasis are obtained by overedge or separate stitches, using absorbable suture material. If the residual cavity is aslope, it is drained, otherwise omentoplasty with drainage is performed.
- Internal transfistulary drainage procedure consists of internal drainage (to the duodenum) of the remaining cavity through a large cysto-biliary fistula. After cleaning, the remaining walls of the cavity are brought together with a series of purse-string or mattress sutures, starting from the bottom and working outward (capitonnage). In the case of a choledechotomy for hydatid material extraction, T tube drainage is performed.

This procedure is indicated for small cysts with a soft pericystium and a large cysto-biliary fistula.

On follow-up, in 20 patients the condition was found to have evolved favorably, but 4 patients developed complications, 3 of whom were treated by medication, but one required surgery. One patient had right focal pneumonia that was treated with antibiotics. One patient had residual cavity suppuration which was treated by antibiotics and percutaneous drainage. An external biliary fistula was observed in 2 cases, in one of which it dried out spontaneously after prolonged drainage. The second case required endoscopic sphincterotomy to reverse the bile flow, with a favorable outcome.

No maternal death occurred in this study, and 21 pregnancies were carried to term and resulted in a live birth. One second-trimester miscarriage occurred, and two cases of fetal distress were noted, which resulted in fetal death.

After regular 36-month follow-up of 14 patients, only one patient relapsed.

## Discussion

The incidence of hydatidosis during pregnancy varies between 1/30,000 and 1/20,000 [1,3]. The hepatic hydatid cyst in pregnant women can give rise to various diagnostic and therapeutic challenges, in making the correct diagnosis and selecting the appropriate procedure and the best time to perform it. The hepatic hydatid cyst can communicate with the bile ducts or with the peritoneal cavity, and it can also become infected. During pregnancy, the risk of the cyst communicating with the bile duct is higher, due to its accelerated growth [4,5]. This is a common and severe complication of hydatidosis.

The clinical presentation varies, ranging from right upper quadrant abdominal pain to acute cholangitis. Abdominal pain is the most frequently encountered symptom. Its usual location is the right upper quadrant, but it can also be epigastric or lower thoracic, or even involve the whole abdomen. Pain was a presenting symptom in 7% to 92% of cases in different studies [6,7]. In our study, abdominal pain was reported in 87.5% of the cases. Fever, which signals a ductal infection, is rarely the first clinical presentation, but it is observed in 45 to 47% of cases [6,8]. In this study 12 patients had fever, a finding compatible with the literature. Vomiting was also reported in the literature, with a frequency of 20.8%, similar to our series. Jaundice often occurs after the abdominal pain and fever. Regardless of its intensity, jaundice is an indicator of common bile duct obstruction. Its frequency varies from 61 to 80% [1,11]. In our study, it was observed in 12 patients (50%). Other symptoms such as hepatomegaly (with a frequency of 14.5 to 75%), a palpable abdominal mass (found in 8 to 50% of cases) have been documented. Communication of the cyst with the bile ducts can present in two ways:

- Acute cholangiitis: In endemic areas, a communicating hydatid cyst is the second most common cause of cholangitis after gallstone. Acute cholangiitis is observed in 48 to 80% of cases of hepatic hydatid cysts, but only 3 (12%) of our pregnant patients had this condition.
- Pain: Pain can be associated with hepatomegaly or with an abdominal mass, and is documented in 18.5 to 22% of cases [11,12]. In our study, pain was reported in 60% of cases.

Blood tests were not specific for diagnosing communicating cysts in our series of pregnant women. The incidence of leukocytosis varies in the literature from 9 to 33.2% of cases [13,14], but was found in 54.2% of our patients. Positive blood cultures vary from 12 to 18.1 of cases [10]. In our patient series, in 6/24 blood culture was positive for Gram negative bacilli.

The frequency of cholestasis and elevated liver enzymes varies among studies, as does the frequency of hepatic failure. Cholestasis was found in 41.7% of our cases and elevated liver enzymes in one half (12 patients). Renal failure, which is a sign of severe sepsis, has been documented in 6.1 to 40% of cases in the literature, but not in any of our patients.

Imaging techniques are paramount in the diagnosis of hydatid cyst communication [15,16]. Abdominal U/S is the technique most often used, and was the only examination performed before surgery in 90% of cases in most studies [17,18]. It visualizes the cysts and determines their type, dimensions, number and location. It allows measurement of the distance between the cyst and the bile ducts and between the cyst and the vascular structures. It also enables detection of latent abdominal injuries [17,19]. Communication between the hydatid cyst and the bile ducts is predominately located in the right liver [10,11], which is, also where most of the non-complicated cysts are found [9,10]. In our patient series, the cyst was located in the right lobe of the liver in 70% of cases.

Increase in cyst size is a major risk factor for rupture. In our study, the size of the hydatid cysts communicating with bile ducts was between 4 cm and 22 cm, with a median size of 10 cm, and 13 cysts (54.2%) were larger than 10 cm in diameter.

In the literature, communication with the biliary tree usually applies to only one cyst [10,20], although two or more cysts may communicate with the bile ducts in the same patient [9,12]. In our series of 24, patients, 5 had two complicated cysts and one had 4, but most (70.8%) had only one complicated cyst.

The hydatid cyst that communicates with the bile ducts is usually old and well developed [21]. Type III and IV cysts are the most common. The communication with the

biliary tree is diagnosed by U/S in 45 to 75% of cases [22]. It is suspected by the image of a sagging cyst, dilatation of the intra- and extrahepatic bile ducts, and the presence of hydatid material in the bile ducts, or by visualization of a communication between the cyst and the bile ducts. These three signs are all combined in 25 % of cases.

A CT scan should only be performed on pregnant women in the third trimester. CT can measure proximity of the cyst to the biliary and vascular structures [23]. A cysto-biliary fistula is suspected by dilatation of the bile ducts close to a loose cystic lesion, and hydatid material can be visualized in the bile duct.

Magnetic resonance imaging (MRI) can be performed after the third month of pregnancy and is more efficient than the CT-scan in detecting complications, especially a communication with bile ducts [37,39]. In this study, all the complicated hepatic hydatid cysts were visualized on U/S and no patient had a CT-scan or an MRI.

A variety of procedures can be followed in the treatment of a complicated hepatic hydatid cyst. The puncture, aspiration, injection, re-aspiration (PAIR) procedure is mostly used for uncomplicated cysts, but its use is not indicated for acute complicated forms due to the frequency of cysto-biliary fistula in these cases. Surgery is the gold standard for treating hepatic hydatid cyst. It is recognized that the second gestational trimester is the best period to perform the surgery as it is associated with a lower risk of miscarriage and premature birth [8]. Although a complicated cyst was not a selection criterion for our patients, most women were operated on during the second trimester. Two fetal deaths and one miscarriage occurred after surgery.

Non-surgical treatment with Albendazole is contraindicated in pregnant women because of its embryotoxic and teratogenic effects demonstrated in animals [9], and it has not been demonstrated to be innocuous in the second and third trimesters [9,11]. For this reason, none of our patients was administered Albendazole.

## Conclusion

The hepatic hydatid cyst is a rare entity in pregnant women. The complicated form presents diagnostic and therapeutic challenge in pregnancy and can be life-threatening to both mother and fetus.

The treatment of a complicated cyst is surgery, at any stage of pregnancy. The maternal and fetal prognosis can be improved by regular monitoring during pregnancy and early detection of the hydatid disease by U/S screening in endemic areas.

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