

CASE SERIES

Acute lithiasis cholangitis in pregnant women: About three cases

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Abstract

Acute lithiasis cholangitis is a rare non-obstetric emergency during pregnancy, which may threaten fetus and mother's life. It requires a codified management in order to avoid complications. In this current study, we aimed to report our center experience in the management of acute lithiasis cholangitis occurring in three pregnant women.

KEYWORDS

acute cholangitis, cholelithiasis, pregnancy

1 | INTRODUCTION

Complicated gallstone diseases are one of the most common causes of non-gynecologic interventions and non-obstetric causes of hospitalization during pregnancy. They are dominated by acute cholecystitis.¹ However, acute lithiasis cholangitis is rare. It constitutes 5% of all complicated gallstone diseases during pregnancy.¹ It is considered as a medico-surgical emergency with an increased morbi-mortality risk for both the mother and the fetus.²

Clinical presentation is variable.³ The association between clinical, biological findings and imaging investigations can make the diagnosis.⁴ The management includes medical with interventional or surgical modalities.

The aim of our study was to describe clinical and morphological features of acute cholangitis in pregnant women, as well as management modalities. We reported all cases of acute cholangitis complicating cholelithiasis in

pregnant women, hospitalized in our surgery department over a period of 8 years from 2014 to 2021.

2 | CASE SUMMARY

2.1 | Clinical presentation

There were three pregnant women without any medical history, with a mean age of 31.3 ± 7 years. Clinical, biological, and imaging features are summarized in [Table 1](#). Two patients had multiple gestations, and one patient was multiparous. The mean term of pregnancy was 19 weeks of amenorrhea. Two patients were in first trimester of pregnancy, and one patient in third trimester.

Patients were referred to our surgery department for abdominal pain in the right hypochondrium evolving for more than 72 h. Fever, nausea, and vomiting were noted in only one patient. Icterus was noted in all patients.

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TABLE 1 Main clinical, biological, and imaging findings for pregnant women with acute cholangitis

	Case 1	Case 2	Case 3
Age	28	40	26
Obstetrical history	G3P1	G3P2	G2P1
Pregnancy term (weeks)	8	12	37
Clinical findings			
Hemodynamic status	Stable	Stable	Stable
Fever	No	No	Yes
Icterus	Yes	Yes	Yes
Abdominal pain	Right hypochondrium	Right hypochondrium	Right hypochondrium
Abdominal examination	tenderness	tenderness	tenderness
Obstetrical examination	Normal	Normal	Normal
Laboratory tests			
WBC (elts/mm ³)	10,000	13,700	12,600
CRP (mg/L)	32	19	27
Renal function	Normal	Normal	Normal
Liver cytolysis	Yes	Yes	Yes
Liver cholestasis	No	Yes	No
Hyperbilirubinemia	Yes	Yes	No
Type of imaging	Only abdominal ultrasound	Only abdominal ultrasound	Only abdominal ultrasound
Imaging findings			
lithiasis gallbladder	Yes	Yes	Yes
Wall-thickness	No	No	No
Gallbladder distension	No	No	No
Bile duct lithiasis	Yes	Yes	Yes
Main bile duct dilation	Yes (17 mm)	No (7 mm)	Yes (11 mm)
Liver abscess	No	No	No
Other abnormalities	No	No	No

Abbreviations: CRP, C-reactive protein; G, gravida; P, pada; WBC, white blood cells.

Abdominal examination revealed tenderness in the right hypochondrium in all patients. There was no palpable abdominal mass.

2.2 | Differential diagnosis, investigations, and treatment

Regarding biologic investigations, the mean white blood count cell was $12,100 \pm 1900$ elements per mm^3 . There was no hyperleukocytosis. The mean C-reactive protein (CRP) was 26 mg/L. One patient had an elevated CRP. Means of aspartate aminotransferase (AST) and alanine aminotransferase (ALT) were, respectively, 84 IU/L and 51 IU/L. Liver cytolysis was noted in all patients. The mean alkaline phosphatase (ALP) was 233 IU/L. Cholestasis



FIGURE 1 Abdominal ultrasound showing dilatation of main bile duct upstream of a stone

was noted in one patient. The mean total bilirubin level was 49.3 $\mu\text{mol/L}$. Two patients had predominantly conjugated hyperbilirubinemia.

Abdominal ultrasound was performed in all patients showing a lithiasis gallbladder that was non-distended and thin walled in all cases. Two patients had a dilated main bile duct with a mean diameter of 11.6 mm. Lithiasis in the common bile duct was observed in all three cases (Figure 1). There was neither hepatic abscess nor liver or pancreas abnormalities.

Abdominal computerizing tomography (CT) scan and biliary magnetic resonance imaging (MRI) have not been performed.

Obstetrical examination and ultrasound were normal in all cases.

Therapeutic modalities and outcomes are summarized in Table 2. All patients received both vitamin K supplementation and antibiotic therapy intravenously. A combination of cefotaxime, metronidazole, and gentamicin was used in all patients with an average time of intake of 9 ± 1.7 days. Tocolysis was indicated in only one patient in third trimester of pregnancy. All patients underwent

surgery. The average time from admission to surgery was one day. The surgical approach was a right subcostal incision in all cases.

Cholecystectomy was performed in all three patients. Cholangiography was performed in two patients showing dilated bile ducts with presence of at least two stones. The number of images was limited to two in both cases. A lead gown was used as fetus protection measures. False biliary fistula was found in one patient allowing direct access, extraction of stones, and verification of main biliary duct with choledochoscope, avoiding cholangiography. Choledocotomy with stone extraction was performed in two patients. Main biliary duct closer with Kehr's T-tube was performed in all patients. There was no reported intraoperative incident.

2.3 | Outcomes and follow-up

Regarding outcomes, postoperative course was simple in all patients. Cholangiography was performed on the seventh postoperative day for two cases showing the vacuity

TABLE 2 Therapeutic modalities and outcomes of pregnant women with acute cholangitis

	Case 1	Case 2	Case 3
Medical management			
Vitamin K	Yes	Yes	Yes
Antibiotic therapy	Cefotaxime, metronidazole, and gentamicin	Cefotaxime, metronidazole, and gentamicin	Cefotaxime, metronidazole, and gentamicin
Tocolysis	No	No	Yes
Surgical modalities			
Time from admission to intervention	1 day	1 day	1 day
Surgical approach	Right subcostal	Right subcostal	Right subcostal
Gallbladder appearance	Sclero-atrophic	Normal	Normal
Biliary fistula	Yes (bilio-biliary)	No	No
Liver abscess	No	No	No
Main bile duct Dilation	Yes	No	Yes
Cholecystectomy	Yes (antegrade)	Yes (antegrade)	Yes (antegrade)
Peroperative Cholangiography ^a	No	Yes	Yes
Number of stones	7 choledochoscope	2	2
Complete stones removal	Yes	Yes	Yes
Biliary duct closer	On Kehr's T-tube	On Kehr's T-tube	On Kehr's T-tube
Postoperative cholangiography via Kehr's T-tube ^a	Yes	Yes	Yes
Kehr's T-tube removal	48 days	42 days	42 days
Outcome	Simple	Simple	Simple
Hospital stay (day)	14	14	12

^aFetus protection measures were also carried out, and the number of images was three.

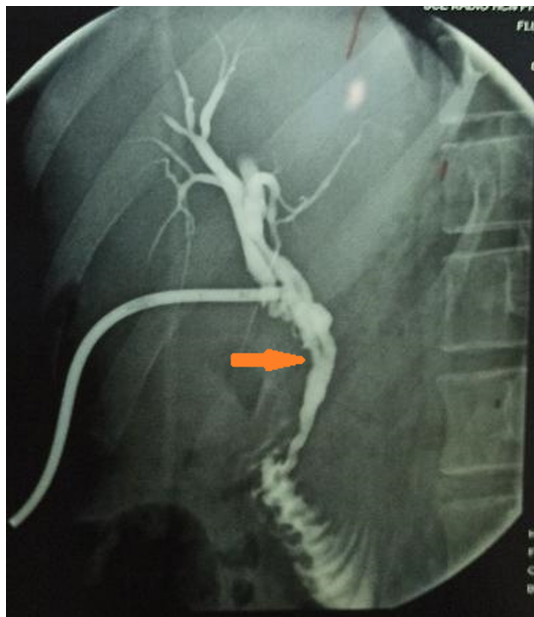


FIGURE 2 Postoperative cholangiography showing bile duct vacuity

of the bile ducts (Figure 2). Fetus protection measures were also carried out, and the number of images was equal to three. The period of hospital stay was 14 days for two patients and 12 days for one patient. Intensive care unit stay was one day for two patients and two days for one patient. There was no complication for both fetus and mother. Pregnancy was carried to term for two patients and one patient was lost to follow-up.

3 | DISCUSSION

We presented our clinical experience concerning the management of acute lithiasis cholangitis during pregnancy. We aimed to describe clinical and morphological features and to report management modalities in order to compare them with literature data.

Our study involved the following main findings. Clinical presentation was the same. The management was multidisciplinary in collaboration with obstetrician-gynecologists. Laparotomy surgery was performed in all cases. Peroperative findings were polymorphous. Simple outcomes were noted for both mother and fetus.

Acute lithiasis cholangitis is rare during pregnancy.¹ Clinical presentation includes the classic symptomatic triad or Charcot's triad, which consists, respectively, of pain, fever, and jaundice over a period of 8–36 h.⁵ As for biologic findings, hyperleukocytosis, positive blood culture, and disturbance liver tests are common.⁶

Nevertheless, it requires the management of both the mother and the fetus because of sepsis' effects.

Imaging investigations are based on abdominal ultrasound as a first-line examination. Biliary MRI and echo-endoscopy may be also useful to detect lithiasis of the main biliary tract bile duct.⁴ MRI is a safe imaging but is not often available for such emergency situation. Instead, echo-endoscopy has a higher sensitivity but is not preferred as first line because of its invasive aspect.⁷

In our case, biliary MRI and magnetic resonance cholangiopancreatography (MRCP) or echo-endoscopy were not performed because of non-availability urgently neither in our hospital nor in our region. CT scan is another non-invasive investigation that may help assessing biliary tree and pancreas. However, CT scan has low sensitivity in the diagnosis of stones in the main bile duct.⁷ Its use is also limited in pregnant women because of radiation-induced teratogenesis.

Obstetrical examination is crucial. It must be done precociously in order to detect fetus complications and to guide the management. This examination should look for gynecologic pathologies that may explain the symptomatology.⁷ Fetal heart rate recording is a simple and rapid examination that allows the assessment of fetal well-being and also allows the detection of uterine contractions.⁷

Obstetric ultrasound is an important element of obstetric assessment. It is used to detect obstetric anomalies as differential diagnosis. It is also essential to assess the fetal well-being. So, endo-vaginal ultrasound can measure the length of the cervix and assess the risk of preterm birth.⁷

Medical therapy modalities of acute cholangitis include filling with fluid and electrolyte rebalancing based on crystalloids. Colloids are contraindicated during pregnancy; only 4% or 5% albumin can be used. Intravenous antibiotic therapy targeting Gram-negative bacteria and anaerobes is obligatory.³ Preceded by blood cultures, it should be empirical, then adapted to the bacteriological results. The choice of molecules must take into account the teratogenic risk for fetus.

Beta-lactams, cephalosporins, and amoxicillin-clavulanic acid can be widely used in combination with an aminoglycoside or metronidazole. These molecules are known to be mildly teratogenic. In order to correct possible hemostasis disorders, the addition of vitamin K is possible without fetal risks.⁸

As for surgical treatment, there are some particularities during pregnancy related to anesthesia, approach, and choice of surgical modalities. The target is to minimize the risk of complications for fetus and to preserve safety for mother. Anesthetic particularities are inherent to the risk of tracheal intubation which is often more difficult with a greater risk of hemorrhage. Good pre-oxygenation with rapid induction sequence is necessary. Drugs' titration

is essential as well as monitoring. Actually, there is no proven risk of teratogenicity due to anesthetic products.⁹

Regarding the approach, both laparotomy and laparoscopy are possible. Various studies have compared these two approaches in terms of prematurity rate or of death in utero. There was not any significant difference between them.⁷ It was proven that laparoscopy can be safe during pregnancy.¹⁰ In our study, only laparotomy approach was used. It was preferred to laparoscopy for several reasons. A biliary MRI has not been performed previously to map lithiasis of the main bile duct. There was no Dormia Basket which is used to retrieve main bile duct stones.¹¹ In our case, cholangiography was performed without preterm delivery or any other fetal complications.

Endoscopic retrograde cholangiopancreatography (ERCP) with sphincterotomy and stone extraction is another therapeutic option for acute cholangitis in pregnant women.¹² It constitutes the treatment choice. It has been shown efficacy and safety in pregnancy for both woman and fetus.¹³ It does not seem to increase risk pregnancy complications.⁶ This modality has not been carried out for our patients because of our limited findings and the non-availability.

Tocolysis, lung maturation with corticosteroids, and transfer to a suitable maternity unit should be discussed according to gestational age.¹⁴

4 | CONCLUSION

Acute lithiasis cholangitis is a rare situation during pregnancy. It constitutes a challenge for surgeon. Diagnosis is often easy considering typical clinical presentation but can lead to serious complications if diagnosis is delayed. In our study, the management was operative using laparotomy with simple outcomes for both mother and fetus. However, the current study had a retrospective design with a small number. Thus, multicenter researches are needed to enhance our findings.

AUTHOR CONTRIBUTIONS

All the authors participated in the design of the study and wrote the manuscript. All authors read and approved the final manuscript. Mohamed Farès Mahjoubi has drafted the work. Anis Ben Dhaou has substantively revised the work. Yasser Karoui, Bochra Rezgoui, and Nada Essid have made substantial contributions to the literature research. Mounir Ben Moussa has made substantial contributions to the conception of the work.

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CONFLICT OF INTEREST

The authors have no conflict of interests.

DATA AVAILABILITY STATEMENT

The data and supportive information are available within the article.

ETHICAL APPROVAL

Our locally appointed ethics committee "Charles Nicolle Hospital local committee" has approved the research protocol and informed consent has been obtained from the subject.

HUMAN AND ANIMAL RIGHTS

Our study complies with the Declaration of Helsinki.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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