

# Choledochal cysts in pregnancy: Case management and literature review

De-Quan Wu, Long-Xian Zheng, Qiu-Shi Wang, Wen-Hua Tan, Shuang-Jiu Hu, Pei-Ling Li

**De-Quan Wu, Long-Xian Zheng, Qiu-Shi Wang**, Department of Surgery, the Second Affiliated Hospital of Harbin Medical University, Harbin 150086, Heilongjiang Province, China

**Wen-Hua Tan, Shuang-Jiu Hu, Pei-Ling Li**, Department of Obstetrics and Gynecology, the Second Affiliated Hospital of Harbin Medical University, Harbin 150086, Heilongjiang Province, China

**Correspondence to:** Professor De-Quan Wu, Department of Surgery, the Second Affiliated Hospital of Harbin Medical University, 157 Baojian Road, City of Harbin, Harbin 150086, Heilongjiang Province, China. zhenglxhrbmu@hotmail.com

**Telephone:** +86-451-86605411

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## Abstract

**AIM:** To evaluate the diagnosis, management principles and long-term results of congenital choledochal cysts in pregnancy.

**METHODS:** Three adult patients were diagnosed as choledochal cysts in pregnancy from 1986 to 1989 and their long-term results were evaluated.

**RESULTS:** The first patient had a Roux-en-Y cysto-jejunosomy with T-tube external drainage and died of septic shock and multi-organ failure 25 d after operation. In the second patient, 4 wk after percutaneous trans-choledochal cyst was drained externally with a catheter under US guidance, four weeks later the patient delivered vaginally, and had a cysto-jejunosomy 3 mo after delivery, and lived well without any complications for 15 years after operation. The third patient received Roux-en-Y cysto-jejunosomy after a vertex delivery by induced labor at 28 wk gestation, and demonstrated repetitively intermittent retrograde cholangitis within 10 years, and then died of well-differentiated congenital cholangioadenocarcinoma one month after re-operation with exploratory biopsy at the age of 36.

**CONCLUSION:** More conservative approaches such as external drainage of choledochal cyst should be considered for pregnant patients with high risk, complete excision of choledochal cyst during hepaticojunosomy or modified hepaticojunosomy is highly recommended at the optimal time.

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## INTRODUCTION

Choledochal cysts in pregnant women represent a diagnostic and therapeutic challenge to a broad spectrum of the medical profession. Not only the rare association, but also the clinical signs and symptoms are obscured by physiological changes that occur during pregnancy. As a result, diagnosis is often delayed until patients present with life-threatening complications. We

reported our experiences in managing three cases of choledochal cyst in pregnant patients.

## MATERIALS AND METHODS

Three adult patients were diagnosed as choledochal cyst in pregnancy from 1986 to 1989 and their long-term results were studied retrospectively (Tables 1, 2).

## RESULTS

A 27-year-old primigravida (patient No.1) at 20-wk gestation presented with jaundice, dark urine, nausea, anorexia, vomiting, intermittent upper abdominal pain, fever and weight loss during the past three months and was admitted to our hospital in February, 1986. Physical examination demonstrated a palpable 12 cm×10 cm mass in the upper right quadrant of the abdomen, a tender protuberant abdomen due to her pregnant state, and fullness in the right upper quadrant. Laboratory evaluation data are shown in Table 2. Ultrasound examination showed a cystic lesion, 12 cm×9 cm×10 cm at the right upper quadrant with a connection to tubular structure, and dilated intrahepatic ducts in both lobes of liver. Impressive diagnosis was gestation with a type IV choledochal cyst. Perioperative fluid therapy and transfusion were administered. A Roux-en-Y cysto-jejunosomy was performed for the choledochal cyst with T-tube external drainage in the proximal part of the cyst. Five hundred milliliter bile was drained from T-tube post-surgery and documented everyday. The patient was afebrile with normal vital signs, flatus was released on d 3 and 10 postoperation. Progressive nausea, vomiting, anorexia, occurred with aggravated jaundice, hypokalemia (2.8 mEq/L), serum electrolyte disturbance and hypoalbuminemia, and then developed to retrograde bile duct infection. The primigravida lost, her consciousness on day 19 postoperation. She was diagnosed as incipient (threatened) abortion, and died of septic shock and multi-organ failure on the 25<sup>th</sup> d postoperation.

Patient No.2 was a 23-year-old primigravida at 36-wk G1P1 gestation with choledochal cyst. She had intermittent upper abdominal pain, progressive nausea, anorexia, vomiting and abdominal tenderness in the upper right quadrant and epigastric burning during the past several weeks. She was admitted to our hospital in February 1987. Physical examination demonstrated a nontender protuberant abdomen due to her pregnant state, and fullness in the right upper quadrant. Her skin and sclera were slightly jaundiced. Sonographic evaluation of the right upper quadrant demonstrated an oval-shaped, 15 cm×16 cm×3 cm cystic mass arising from the portal hepatic separated from the gallbladder, and diffuse dilatation of intrahepatic left ductal system. Clinical diagnosis was a type IV choledochal cyst with gestation (G1P1). Laboratory evaluation data are shown in Table 2. Percutaneous trans-choledochal cyst operation was performed on the patient with a catheter placed for external drainage under the abdominal ultrasonography guidance. About 3000 mL sap green bile was excreted from the cyst, and then about 600 mL sap green bile was excreted daily. Gradually, her clinical conditions were improved and remained stable and her pregnancy progressed without problems. She delivered vaginally 4 wk later without

**Table 1** Information in 3 patients diagnosed as choledochal cysts in pregnancy

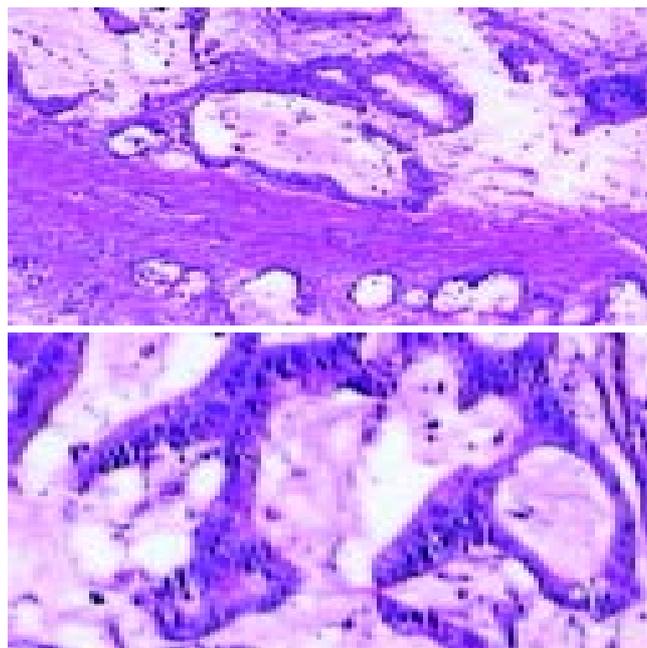
Patients No.	Age(yr)	Type	Size (cm)	Chief Complaints	Treatment		Complication		Survival time (postoperation)	Prognosis (postoperation)
					Initial operation	Final operation	Early	Late		
1	27	IV	12×9×10	Abdominal pain Jaundice Abdominal mass	Roux-en-Y cystojejunostomy with T-tube external drainage		Septic shock and multi-organ failure	25 d	died	
2	23	IV	15×16×3	Abdominal pain Jaundice	Plasement and fixation of a catheterization	Roux-en-Y cystojejunostomy		15 yr	survived	
3	26	I	20×20×20	Jaundice Abdominal mass	Roux-en-Y cystojejunostomy	Exploratory biopsy	Retrograde cholangitis Intermittent recurrence of Retrograde cholangitis Cholangioadenocarcinoma	10 yr	died	

**Table 2** Analysis of pre-operation laboratory values for 3 patients

Parameters	Normal range	Patient No. 1	Patient No. 2	Patient No. 3
Total bilirubin (μmol/L)	1.6-20.6	59	57	26
Direct bilirubin (μmol/L)	0-6.4	8.0	8.7	8.1
Alkaline phosphatase (U/L)	35-150	171	63	110
r-GT (U/L)	0-60	51	58	26
Aspartate aminotranferase (U/L)	0-40	37	37	37
Alanine aminotranferase (U/L)	0-40	5	11	102
Total protein (g/L)	60-85	40	67	55
Albumin (g/L)	35-55	18	39	32
Globin (g/L)	-	22	25	23
Potassium (mmol/L)	3.5-5.5	1.9	4.2	4
Sodium (mmol/L)	135-145	134	128	139
Chloride (mmol/L)	101-111	89.3	96.9	105
Hemoglobin (g/L)	120-160	74	150	122
Erythrocyte (10 <sup>12</sup> /L)	3.5-5.5	2.4	4.81	3.9
Hemoleukocyte (10 <sup>9</sup> /L)	3.5-10	5.8	4.4	9.7
Platelet (10 <sup>9</sup> /L)	100-300	92	177	128

complications. As choledochal cyst was extensively attached to the surrounding tissues, it was dissected using cystojejunostomy 3 mo after delivery. Choledochal cyst was histologically confirmed. The patient was discharged from hospital 40 d postoperation and was well without any complications for 15 years.

Patient No.3, a 26-year-old multiparous woman at 28-wk gestation with choledochal cyst was admitted to our hospital in July 1989. Her chief complaint was jaundice and she had a palpable mass in the upper right quadrant of the abdomen during the past 20 d. On examination, the patient was afebrile with normal vital signs with skin and her sclera slightly jaundiced. She had a soft palpable 8 cm×10 cm mass in the upper right quadrant of the abdomen without tenderness. Laboratory evaluation data are shown in Table 2. Ultrasound examination showed a large cyst of 20 cm×20 cm×20 cm at the right upper quadrant connected to tubular structure. The patient gave a vertex delivery by induced labor at 28-wk gestation on the second day of admission. Following treatment with prophylactic antibiotics, the patient received Roux-en-Y cystojejunostomy on d 8 after delivery. The patient had 4 or 5 times of recurrence of intermittent retrograde cholangitis annually for 10 years, lasting for about 20 d each time, and relieved after a few days of conservative antibiotics. However, the patient was not re-operated due to the extensive adherence with surrounding tissues, and died of well differentiated congenital mucinous cholangioadenocarcinoma proved at the age of 36 years.



**Figure 1** High-columnar or flat carcinoma cells with darkly stained nuclei and variation in size and shape arranged in irregular glandular pattern with intra- and extra-glandular mucin (hematoxylin and eosin, 40×10).

## DISCUSSION

Choledochal cyst is a focal dilatation of the biliary tract. Although rare in adulthood, it is diagnosed more frequently with the advances in biliary imaging techniques<sup>[1]</sup>. Of the numerous descriptions and classifications of choledochal cyst, the most practical is the classification proposed by Alonso-Lej 1 and modified by Todani<sup>[1]</sup> in 1977 (Table 3). The major point of this classification is that the gallbladder is not usually distended and arises from a small cystic duct, not at the upper end of the cyst<sup>[2]</sup>.

**Table 3** Todani modification of the Alonso-Lej, classification system of choledochal cyst

Type I	Fusiform dilation of the extrahepatic bile duct
Type II	Single saccular dilation or diverticulum of the extrahepatic bile duct
Type III	Dilation of the intraduodenal portion of the bile duct
Type IVa	Combined intra-and extrahepatic dilation of the bile duct
Type IVb	Multiple dilations of the extrahepatic bile duct
Type V	Isolated or diffuse intrahepatic biliary dilation (Caroli's disease when associated with hepatic fibrosis)

Since the first pathological description of a choledochal cyst by Vater in 1723 and the first clinical report by Douglas in 1852, the aetiology of this abnormality has been controversial<sup>[3]</sup>. It is unclear whether choledochal cyst is congenital, or acquired. In 1936 Yotsuyanagi<sup>[3]</sup> suggested that choledochal cysts arose from inequality in the vacuolization of the biliary tract in early embryonic life. The common channel theory proposed by Babbitt<sup>[3]</sup> in 1969 is most widely accepted, which was based on an abnormality of the pancreaticobiliary junction and the formation of an abnormally long common channel (greater than 15 mm) outside the control of the sphincters of Boyden. This configuration permits pancreatic enzymes to reflux into the common bile duct. The pancreatic enzymes then lead to persistent inflammation, epithelial denudation, thinning of the bile duct wall and distal obstruction, then eventually cyst formation. The incidence of choledochal cysts was reported as 1 in 13 000 to 1 in 2 million patients<sup>[3,4]</sup>. It affects females 4 times as often as males, and more common in Asians and presents mainly in infancy and childhood (60%)<sup>[1]</sup>. It is necessary to classify the type of cysts and to recognize the presence of an abnormal pancreaticobiliary duct junction, visualization of both the biliary tree and pancreatic duct. Thus, direct cholangiography, especially ERCP, is beneficial. But intraoperative cholangiography is not sensitive enough to detect the presence of an abnormal pancreaticobiliary duct junction. Radiographic visualization of both the biliary tree and pancreatic duct prior to surgery is helpful for surgical manipulation and complete excision of the cyst<sup>[5-8]</sup>. Other pathologic features of choledochal cysts include acute and chronic mucosal inflammation, mucosal dysplasia, and absence of smooth or elastic fibers. A true mucosal lining may be hard to find, or it is usually cuboidal or columnar and frequently ulcerated if it is present. The cyst wall varies from 1 to 10 mm in thickness. Mucus-producing glands are rarely seen. There is usually a large amount of fibrosis, some of which may be involved in luminal stenoses. The bile is often very thick, and sometimes less pigmented than normal. Biliary calculi are uncommon. Pathologic complications include biliary obstruction, cholangitis, hepatic abscess, rupture, or development of cancer. Cholelithiasis due to choledochal cysts is unusual<sup>[2,9]</sup>.

Clinical manifestations are nonspecific and variable. The most common symptoms are abdominal pain and jaundice. If untreated, the condition may be fatal due to ascending cholangitis, biliary cirrhosis or diffuse peritonitis following

rupture of the cyst. Malignancy might also develop in the cyst and occur in pregnant women<sup>[10]</sup>. Choledochal cyst in pregnancy is rare but poses a threat to both the mother and fetus. The maternal complications include cholangitis, pancreatitis, peritonitis and even malignancy. The fetal complications include fetal loss and preterm labor. Most reported cases were diagnosed when women presented with symptoms. It has been suggested that pregnancy may exacerbate the symptoms due to hormonal effect, compression by the gravid uterus and increase in intraabdominal pressure during pregnancy and postpartum. The cyst might be asymptomatic during the first pregnancy<sup>[11,12]</sup>, as demonstrated in our third patient. Asymptomatic cysts discovered on routine US could be observed with serial US examinations<sup>[11]</sup>.

Diagnosis of choledochal cysts during pregnancy is difficult. Radiologic imaging, ultrasound, computed tomography, cholangiography and biliary scintigraphy, could clinch the diagnosis<sup>[1,12-15]</sup>. Although ultrasonography is a common useful investigation, difficulty may arise during pregnancy due to distortion of the normal abdominal anatomy and gravid uterus. Furthermore, ultrasound examination cannot demonstrate anatomic details of the biliary tree. Due to exposure of the fetus to ionizing radiation, contrast and ionizing imagings, such as CT or ERCP, should be avoided in pregnancy<sup>[11,12,16]</sup>. Magnetic resonance imaging (MRI) could provide clear visualization of the relations between the choledochal cyst and biliary tree and of the extent and size of the choledochal cyst. Therefore, MRI is the investigation of choice in doubtful cases<sup>[12,17]</sup>. To improve the management of this potentially serious condition in pregnancy, both clinicians and radiologists should be aware of this possibility in women with a right upper quadrant mass.

Concerning the optimal time of treatment, conservative management is commonly adopted during pregnancy. Once the diagnosis has been established, surgery is the only option for treatment<sup>[11,12]</sup>. Surgical time is critical during pregnancy and the operative risk to the fetus and mother has to be balanced against the likelihood of cyst-related complications<sup>[11,12]</sup>. Any patient who presents with a symptomatic or rapidly enlarging choledochal cyst or with cyst-related complications during pregnancy should undergo urgent treatment. Unfortunately, surgery during pregnancy has been associated with high fetal and maternal morbidity and mortality rate<sup>[1,11,12,15]</sup>. Therefore, other more conservative approaches might have to be adopted until surgery could be performed under optimal conditions. Percutaneous cyst decompression might be done to relieve symptoms of pain and jaundice<sup>[1,11,12,15,16]</sup>. In addition, antibiotics should be administered for treatment of cholangitis or for long-term prophylaxis, and the need for subsequent definitive cyst surgery should not be obviated<sup>[11,12]</sup>. An operation should ideally be performed in the second trimester when the risk of surgery and anesthesia is lowest. When symptoms occur in the first trimester, surgery should be postponed unless the life of the mother is in danger, while in the third trimester, early cesarean section should be performed when amniocentesis indicates that the fetus is sufficiently mature. At the same time, a temporizing or definitive cyst operation could be undertaken as necessary<sup>[1,11,12]</sup>. Definitive surgery depends on the cyst type and associated hepatobiliary pathology. Definitive surgical excision of the cyst can avoid postpartum complication and non-cyst excision may induce long-term complications.

Early reports suggested that internal or external drainage of a choledochal cyst by choledochocystojejunostomy or T-tube choledochocystostomy was a satisfactory treatment, but with a longer follow-up it has become clear that complications such as suppurative cholangitis, lithiasis, pancreatitis, secondary biliary cirrhosis, portal hypertension and intrahepatic abscess occurred in up to 40 percent of cases<sup>[3,4,9,15]</sup>. This is similar as our results. The increased risk of bile duct carcinoma in

choledochal cysts has been well characterized<sup>[3,4,9,10,14,15]</sup>. The reported incidence of biliary tract carcinoma in choledochal cysts varied from 2.5% to 17.5%, significantly higher than that found in the general population, which was from 0.01% to 0.05%<sup>[3,4,9,10,14,15]</sup>. The incidence of cancer in patients with a choledochal cyst and in those undergone enteric drainage without cyst excision was much higher than that of carcinoma of the bile ducts in the general population. The age-related incidence of cyst-associated cancer has been shown to increase from 0.7 % in the first decade of life to 14.3 % after 20 years of age. It means the favorable outcome in congenital choledochal cyst patients was due to earlier diagnosis<sup>[3,4,9,10,14,15]</sup>.

Kasai *et al.*<sup>[3]</sup> reported firstly the increased incidence of carcinoma in choledochal cysts and advocated primary cyst excision. The advances in diagnostic and therapeutic procedures and increased operative experience have lowered the mortality rate to 0-7%<sup>[3,4,9,10,14,15]</sup>. Roux-en-Y choledochojejunostomy has replaced choledochoduodenostomy as the preferred operative procedure because of the high morbidity rate of cholangitis and the frequent need for reoperation later<sup>[3,4,9,15]</sup>.

Excision of types I, II and IV choledochal cysts is now widely accepted because of the lower incidence of postoperative complications. In contrast to cyst enterostomy, cyst excision with hepaticojejunostomy had satisfactory results. Although the occurrence of intrahepatic cholangiocarcinoma after the excision of a type I cyst has been reported<sup>[18,19]</sup>, but cyst excision is the primary choice of treatment for type I cysts. Type III cysts require adequate drainage and generally can be managed by endoscopic sphincterotomy and cannot be undertaken, operative sphincteroplasty with transduodenal cyst excision might be attempted<sup>[5]</sup>. Sphincteroplasty either endoscopically or surgically has been found to be satisfactory<sup>[20]</sup>. Treatment for type IV cysts is still controversial. Either excision of the extrahepatic cyst alone<sup>[5]</sup> or total cyst excision including hepatectomy<sup>[21]</sup> has been recommended. Concerning type V cysts, some authors recommended hepatic resection for unilobar Caroli's disease<sup>[1]</sup>.

Complete excision of the extrahepatic bile duct from the hepatic hilum to the pancreaticobiliary duct junction has become the choice of treatment for types I and IV cysts<sup>[3-5,9,11,19,21-23]</sup>. However, it should be mentioned that since complete excision seems to be difficult in some patients, pancreaticoduodenectomy or hepatic resection should then be considered. In such cases, the distal choledochus is resected just above the pancreaticobiliary duct junction with the aid of preoperative ERCP and intraoperative US to avoid injuring the pancreatic duct. In the hepatic hilum, the hepatic duct must be resected at the hilum, and hepaticojejunostomy with a wide opening by plastic of both the hepatic ducts is necessary.

The reason why the three patients turned out quite different was their different clinical conditions. Our first Patient's situation was not quite good in peri-operation, she had a progressive nausea, vomiting, anorexia, fever, weight loss, and anemia with skin and her sclera slightly jaundiced, hypoalbuminemia, as well as disturbed blood electrolytes. Although the patient received fluid therapy and transfusion in peri-operation, the volume of blood was not enough to improve her clinical conditions within a short period of time, such as anemia, hypoalbuminemia. Gentamicin sulfate 240 000 iu one time per day could not prevent occurrence of retrograde bile duct infection, and the patient died of septic shock and multi-organ failure on day 25 postoperation.

Choledochal cyst in our No. 2 patient was drained externally with a catheter under the abdominal ultrasonography guidance, and she delivered safely. As the choledochal cyst was extensively infused with surrounding tissues, a cysto-jejunosomy was performed 3 mo after delivery. The patient's general condition was good. In our study, the No.3 patient developed a malignant tumor, because she did not receive re-operation for complete cyst excision, hepatojejunosomy or modified hepatojejunosomy

for treatment of repeated occurrence of intermittent retrograde cholangitis, which could be treated or avoided by early diagnosis and early complete cyst excision<sup>[3,4,9,10,14,15]</sup>, which are very important for the treatment of congenital choledochal cyst patients. Although cyst excision did not completely eliminate the risk of intrahepatic cholangiocarcinoma after the excision of a type I cyst<sup>[3-5,9-11,14,15,18-22]</sup>, but it significantly decreased the incidence of bile duct carcinoma. Thus, laparotomy during pregnancy should be avoided if possible and non-cyst excision cases should be explored to remove the cyst completely in the optimal surgical time.

Although choledochal cysts rarely occur in pregnancy, clinicians need to be aware of the condition. Inappropriate therapy may be catastrophic for both mother and child. This is why we recommend that patients should be admitted to a specialized hospital once the diagnosis is established. Complete excision of choledochal cyst with Roux-en-Y hepaticojejunostomy or modified Roux-en-Y hepaticojejunostomy is the choice of treatment for types I, II and IV choledochal cysts in non-pregnant adult patients; whereas in pregnancy, a more conservative approach should be adopted until the time when the surgical risk is lowest. Percutaneous external drainage of choledochal cyst under the abdominal ultrasonography guidance is indicated, if a complication of cystic rupture occurs. Complete excision of choledochal cyst with hepaticojejunostomy or modified hepaticojejunostomy is helpful in avoiding late complications such as retrograde cholangitis, biliary tract carcinoma, and is the most ideal choice of treatment for types I, II, IV choledochal cysts in pregnant adult patients, and is therefore highly recommended.

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