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## **Congenital hepatic cyst: Eleven case reports**

Du *et al.* Infant hepatic cyst

Chun-Xia Du, Chang-Gui Lu, Wei Li, Wei-Bing Tang

### **Abstract**

#### **BACKGROUND**

Liver cysts in infants are uncommon. With modern diagnostic imaging, we can achieve an early diagnosis of congenital hepatic cysts. Our purpose was to investigate the clinical features, surgical treatment methods and prognosis of infants with congenital hepatic cysts. Herein, we report a case series of congenital hepatic cysts.

#### **CASE SUMMARY**

Eleven infants with hepatic cysts were retrospectively analysed. Ten of them had simple hepatic cysts, and a girl with a large hepatic mass was diagnosed with a solitary intrahepatic biliary cyst accompanied by a choledochal cyst. Among the ten simple hepatic cysts, eight were solitary and two were multiple. A total of 87.5% (7 of 8) of infants with solitary hepatic cysts were detected before delivery, and 86% (6 of 7) of those cysts were located in the right lobe of the liver. Surgical intervention was required for symptomatic hepatic cysts. Cyst resection or unroofing with fulguration of the cyst bed was employed. No recurrence of cysts was observed in these infants.

#### **CONCLUSION**

Congenital hepatic cyst is a condition with a narrow differential diagnosis. Accurate diagnosis is essential for appropriate management. Unroofing is the favoured treatment in infants with symptomatic cysts. Most infants with congenital hepatic cysts have a good prognosis.

**Key Words:** Hepatic cyst; Infant; Surgery; Case series; Case report

Du CX, Lu CG, Li W, Tang WB. Congenital hepatic cyst: Eleven case reports. *World J Clin Cases* 2022; In press

**Core Tip:** <sup>2</sup> Liver cysts in infants often pose significant diagnostic challenges, and paediatric surgeons should be aware of the characteristics of liver cysts to ensure appropriate treatment. Here, we report our experience with congenital hepatic cysts in infants, including a case of solitary intrahepatic biliary cyst, and review the recent literature.

## **INTRODUCTION**

Congenital hepatic cysts are comparatively rare, but with the routine use of prenatal ultrasound, liver cysts are being diagnosed earlier and more often. <sup>2</sup> Liver cysts in newborns <sup>2</sup> often pose significant diagnostic challenges. Solitary liver cysts can be further subdivided as simple liver cysts (SLCs) and solitary intrahepatic biliary cysts (SIBCs), depending on the biliary connection<sup>[1,2]</sup>. Some SLCs are symptomatic in childhood, even in newborns<sup>[3]</sup>, while they are commonly found in adults incidentally. It is imperative to be aware of the characteristics of liver cysts in infants to ensure that any intervention is appropriate.

Some paediatric congenital hepatic cysts have been reported, but most descriptions have been case reports. <sup>1</sup> Here, we report a series of eleven infant patients diagnosed with hepatic cysts at our department whose clinical features and surgical treatments are described in detail. We discuss the possible aetiological basis and review the common manifestations and surgical options for congenital hepatic cysts in infants.

## **CASE PRESENTATION**

### ***Chief complaints***

Case 1: A <sup>12</sup> 1-mo-old girl presented with a prenatal cystic abdominal mass at 34 wk of gestation; Case 2: A 1-mo-old boy presented with a prenatal cystic abdominal mass at 26

wk of gestation; Case 3: A 13-h-old boy presented with a prenatal cystic abdominal mass at 38 wk of gestation; Case 4: A 4-mo-old girl presented with a prenatal cystic abdominal mass at 28 wk of gestation; Case 5: A 6-d-old boy presented with a prenatal cystic abdominal mass at 39 wk of gestation; Case 6: A 1-mo-old girl presented with a prenatal cystic abdominal mass at 24 wk of gestation; Case 7: A 1-mo-old boy presented with a prenatal cystic abdominal mass at 26 wk of gestation; Case 8: A 3-h-old boy presented with an abnormal umbilical cord for 3 h; Case 9: A 2-mo-old boy presented with a prenatal cystic abdominal mass at 26 wk of gestation; Case 10: A 1-h-old boy presented with an abnormal umbilical cord for 1 h; Case 11: A 1-mo-old girl presented with abdominal distension and an abdominal mass for 3 d.

### *History of present illness*

**Case 1:** The patient remained asymptomatic.

**Case 2:** The patient suffered vomiting.

**Case 3:** The patient remained asymptomatic, but progressive enlargement of the cyst was observed.

**Case 4:** The patient remained asymptomatic.

**Case 5:** The patient suffered abdominal distension.

**Case 6:** The patient remained asymptomatic, but progressive enlargement of the cyst was observed.

**Case 7:** The patient suffered abdominal distension and vomiting.

**Case 8:** Emergency surgery for omphalocele was conducted when the patient was born.

**Case 9:** The patient was born at the 34 + 2<sup>nd</sup> gestational week. The patient remained asymptomatic, but progressive cyst enlargement was observed.

**Case 10:** The patient was born at the 33 + 4<sup>th</sup> gestational week, and emergency surgery was conducted for omphalocele.

**Case 11:** The patient was premature with a birthweight of 1850 g. When the girl was born, she <sup>2</sup> was admitted to the neonatal intensive care unit for evaluation and management. The infant suffered from respiratory problems as a result of progressive abdominal distension.

#### <sup>1</sup> *History of past illness*

None of the patients had a history of specific illnesses.

#### *Personal and family history*

None of the patients had a relevant personal or family history.

#### *Physical examination*

**Cases 1 and 3:** There was slight jaundice upon admission.

**Cases 2, 3, and 9:** A mass was found in the abdomen.

**Cases 8 and 10:** A lump in the umbilical cord surrounded by a capsule was found when they were admitted.

**Cases 5 and 7:** Physical examination revealed a distended abdomen.

**Case 11:** The patient experienced abdominal distension and breathing difficulty when she was admitted, with slight jaundice.

**Cases 4 and 6:** No positive signs were found.

#### *Laboratory examinations*

**Case 1:** Alanine aminotransferase, 15 U/L; aspartate aminotransferase, 28 U/L; total bilirubin, 46.67  $\mu\text{mol/L}$ ; and direct bilirubin, 35.94  $\mu\text{mol/L}$ .

**Case 3:** Alanine aminotransferase, 13 U/L; aspartate aminotransferase, 43 U/L; total bilirubin, 146  $\mu\text{mol/L}$ ; and direct bilirubin, 132.4  $\mu\text{mol/L}$ .

**Case 9:** Alanine aminotransferase, 8 U/L; aspartate aminotransferase, 161 U/L; total bilirubin, 42.23  $\mu\text{mol/L}$ ; and direct bilirubin, 27.87  $\mu\text{mol/L}$ .

**Case 11:** White cell count,  $12.99 \times 10^9/\text{L}$ ; C-reactive protein, 28 mg/L; total bilirubin, 35.11  $\mu\text{mol/L}$ ; direct bilirubin 21.99  $\mu\text{mol/L}$ .

**Cases 2, 4, 5, 6, 7, 8, and 10:** Liver function and bilirubin showed no abnormal changes.

#### *Imaging examinations*

**Case 1:** Ultrasound showed a mass (41 mm  $\times$  35 mm) in the right upper abdomen when he was admitted. Computed tomography (CT) was then performed.

**Case 2:** Ultrasound showed a mass (75 mm  $\times$  50 mm  $\times$  63 mm) in the right upper abdomen when he was admitted. Upper gastrointestinal examination (UGI) showed that the bowel was compressed.

**Case 3:** Ultrasound showed a mass (64 mm × 34 mm) in the middle upper abdomen when he was admitted. Magnetic resonance imaging (MRI) showed that the abdominal organs were compressed.

**Case 4:** Ultrasound and MRI showed a mass (40 mm × 23 mm) in the right upper abdomen when he was admitted.

**Case 5:** Ultrasound showed a mass (60 mm × 50 mm × 40 mm) in the right upper abdomen when he was admitted. CT showed that the bowel was compressed.

**Case 6:** Ultrasound and CT showed a mass (39 mm × 25 mm) in the right upper abdomen when he was admitted.

**Case 7:** Ultrasound and CT showed a mass (51 mm × 35 mm) in the right upper abdomen when he was admitted.

**Case 9:** An ultrasound scan showed a 6.9 cm × 5.6 cm cystic mass in the right upper abdomen and an intrahepatic cyst 2.3 cm × 1.2 cm in size, with no intrahepatic biliary duct dilatation.

**1** MRI showed a mass (3.0 cm × 2.3 cm × 2.5 cm) in the right middle cranial fossa and a mass (0.5 cm × 0.5 cm × 0.5 cm) in the sellar region. Specifically, this patient had comorbid polycystic kidneys diagnosed on the first ultrasound scan.

**Case 11:** An ultrasound scan (Figure 1A) showed a mass in the abdomen. CT scans (Figure 1B) showed two large intraabdominal cysts. The larger cyst (8.5 cm × 7.9 cm) was located in the right lobe of the liver, and the other cyst (4.3 cm × 2.0 cm) located in the porta hepatis was considered a choledochal cyst. The extrahepatic bile ducts and gallbladder were obscured by the massively enlarged cyst. No definite intrahepatic bile duct dilatation was observed. However, compression effects on other organs were noticeable.

**Cases 8 and 10:** Since the cysts were discovered accidentally during surgery, no imaging data were available.

### **FINAL DIAGNOSIS**

#### ***Cases 1, 2, 3, 4, 5, 6, 7, and 9***

Combined with the patient's medical history, the final diagnosis was a SLC.

#### ***Cases 8 and 10***

The final diagnosis was omphalocele accompanied by a SLC.

#### ***Case 11***

An uncertainly diagnosed mass accompanied by a choledochal cyst was considered before surgery. Combined with the intraoperative findings, the final diagnosis was a solitary intrahepatic biliary cyst (SIBC).

### **TREATMENT**

All patients underwent surgery. Total cyst excision was performed for four patients, and unroofing with fulguration of the cyst bed was performed for three patients. The cysts were operated on *via* a laparoscopic approach in three patients. Open abdominal excision or fenestration was conducted in four patients. The reasons for converting to open laparoscopy were uncertain diagnosis, abdominal adhesion or large size. Details of the clinical and pathological presentations of the cases are shown in Tables 1 and 2. A postoperative pathology test identified all cysts as SLCs. In regard to size, the solitary hepatic cysts averaged 5.43 cm in diameter, larger than that of multiple cysts. Six of seven solitary cysts were located in the right lobe of the liver. The simple hepatic cysts in this series were lined by cuboidal epithelium that stained positively for cytokeratin. The fluid from the simple cysts was yellow or yellowish.



For Case 11, an uncertainly diagnosed mass accompanied by a choledochal cyst was considered; therefore, we performed a laparoscopic examination, revealing obvious adhesion and a large cystic mass in the abdomen. The gallbladder was identified. The cyst wall was opened, and it was filled with bile-stained fluid and muddy stones, so an intracavitary catheter was placed. She was presumed to have SIBC without pathologic examination.

### **OUTCOME AND FOLLOW-UP**

No recurrence of cysts was observed in any of these infants.

For case 9, the cysts in the kidneys spontaneously and completely resolved after two weeks.

For case 11, after the operation, the abdominal distension and breathing difficulty were relieved. Total bilirubin was 17.87  $\mu\text{mol/L}$  with a direct bilirubin of 10.72  $\mu\text{mol/L}$  on postoperative day 6. Bile drainage through the catheter stopped on the ninth day after the operation, but the patient developed fever, abdominal distension and tense abdominal skin. Blockage of the drainage tube was considered and confirmed by emergency operation. After adjusting the drainage tube, the infant had an uneventful postoperative course. The bile drainage decreased and turned clear before it eventually stopped. Ultrasonography confirmed the complete disappearance of the intrahepatic cyst. The drains were removed two months and five days after the initial operation. When the patient underwent surgery for the choledochal cysts at nine months of age, no hepatic cysts were observed. A follow-up ultrasound examination showed no recurrence when she was one year and one month old.

### **DISCUSSION**

Congenital liver cysts are categorized as nonparasitic simple cysts or polycystic liver disease (PCLD). Simple hepatic cysts can be solitary or multiple<sup>[4]</sup>. Although multiple cysts in the liver are common in these patients, the condition is distinct from PCLD, an autosomal dominant condition associated with PKD1 and PKD2 gene mutations<sup>[5]</sup>.

PCLD may be associated with congenital polycystic kidney disease or dilatation of the bile ducts (Caroli's disease), while SLCs are usually never accompanied by other organ cysts<sup>[6]</sup>. However, in our series, our 9<sup>th</sup> patient was diagnosed with SLCs complicated with polycystic disease of the kidney. This is not consistent with a previous study. Perhaps further gene screening of this infant can provide a definitive explanation.

4  
18 The pathogenesis of hepatic cysts is related to the type of cyst. It has been hypothesized that SLC might be caused by abnormal bile duct formation during embryogenesis<sup>[7]</sup>. SIBC is believed to arise from inadequate drainage of part of the intrahepatic biliary tree<sup>[3,8]</sup>. Most solitary hepatic cysts are SLCs. Pathologically, SLC has a single layer of cuboidal or columnar epithelium lining the inside of the cyst. The cystic fluid may be clear<sup>[9]</sup>. SIBC displays the same characteristics as SLC, but it contains bile<sup>[1]</sup>. We reported a case diagnosed with SIBC based on previous reports<sup>[1,8,10]</sup>. Unfortunately, retrograde cholangiography could not be performed because of a wide adhesion in the abdomen. The boy with SIBC reported by Soyer *et al*<sup>[8]</sup> was revealed by prenatal ultrasonography. He suffered respiratory distress, similar to the premature girl in our report. Additionally, our reported girl exhibited abdominal distension and jaundice. The cyst was not found prenatally. Moreover, the hepatic cyst was accompanied by a choledochal cyst in this report. Interestingly, the previously reported boy has an older brother who was diagnosed with a choledochal cyst. These findings may indicate an aetiological relationship between SIBC and choledochal cysts.

Solitary liver cysts are slightly more common in female patients, with a 1.5:1.0 female-to-male ratio. Previous research has shown that oestrogen may account for this sex difference<sup>[11]</sup>. In contrast to previous reports, more boys exhibited congenital hepatic cysts in this study. This might be due to the small sample size resulting in random variation, and multicentre studies with more samples will be more credible in establishing the sex ratio.

In our cases, the median gestational age at detection for prenatally detected cysts was 30 wk (24-39 wk). These infants with solitary hepatic cysts were all born at term. An ultrasound scan was postnatally performed on the infants with prenatally detected

cysts. Asymptomatic hepatic cysts can be monitored by ultrasound because a complete spontaneous resolution of prenatally diagnosed hepatic cysts has been reported in a recent systematic review and meta-analysis<sup>[12-14]</sup>. Occasionally, an intraoperatively encountered cyst is found to be pedunculated and is easily excised. A simple cyst resection is recommended for these cysts due to the risk of torsion.

Nevertheless, simple cysts can become symptomatic cysts requiring surgical treatment when progressive enlargement or symptoms are present or if the imaging characteristics cannot lead to a definitive diagnosis. Large simple cysts in newborns may cause abdominal distention, pain, jaundice, feeding difficulties and respiratory distress<sup>[15-17]</sup>. If it is over 5 cm and/or the cyst has radiologic findings that are suspicious for neoplasia<sup>[11]</sup>, surgery becomes necessary.

Ultrasonography-guided percutaneous aspiration may serve as a diagnostic tool, with a recurrence rate of 100%<sup>[18]</sup>. To achieve permanent ablation, instillation of a sclerosant, such as ethanol, tetracycline, minocycline or pantopaque, is conducted. The sclerosing agent induces an obvious inflammatory reaction that potentially produces obliteration of the cyst space<sup>[19,20]</sup>. Communication of the cyst with the biliary tree is an absolute contraindication to injection sclerotherapy<sup>[21]</sup>.

Most authors advocate surgical treatment for symptomatic hepatic cysts, including unroofing, cyst excision or liver resection<sup>[22,23]</sup>. Total excision should be attempted for easily accessible SLCs because it eliminates the risk of recurrence<sup>[24]</sup>. In most cases, complete excision is not possible because of the close proximity of the cyst wall to nearby parenchymal and hilar structures, and a portion of the cyst wall should be excised.

Unroofing liver cysts can be undertaken as an open or laparoscopic surgery<sup>[25]</sup>. Laparoscopic unroofing has become the gold standard treatment with low recurrence rates between 0% and 13.8%<sup>[26,27]</sup>. Some centres employ fulguration of the cyst bed and omental transposition to reduce the risk of cyst recurrence<sup>[28]</sup>. However, recent data suggest that transposition of the omentum is not needed. The greater omentum flap could contribute to local recurrence by reducing the size of the parenchymal opening

and inducing adhesion<sup>[29]</sup>. Recently, laparoscopic liver cyst fenestration with indocyanine green (ICG) fluorescent imaging has been believed to be particularly useful for delineating adequate margins for the procedure and for avoiding duct injuries<sup>[30-32]</sup>. In this study, unroofing, followed mainly by fulguration of the cyst bed, led to no recurrence.

Hepatic lobectomy for simple cysts has been reported in patients with complex or recurrent cystic disease<sup>[33]</sup>. Additionally, for symptomatic SIBC caused by insufficient drainage during the neonatal period, internal drainage *via* cystoenterostomy or Roux-en-Y hepaticojejunostomy is recommended<sup>[34]</sup>. However, partial excision of symptomatic SIBC also had a good prognosis in a previous study<sup>[8]</sup>. Recently, a case reported that endoscopic ultrasound-guided drainage could also achieve a good prognosis<sup>[35]</sup>. This is consistent with our experience. In our opinion, the excellent prognosis of our patient is potentially associated with bile duct inflammation, which induced the occlusion of the small canalicular bile ducts. A long-term follow-up is certainly necessary.

With the widespread use of prenatal sonography, aspiration of giant hepatic cysts in the foetus in utero has been reported<sup>[34,36]</sup>. Due to fairly high recurrence rates in adults, simple aspiration of the cyst should be performed only for the emergency control of symptoms.

Overall, congenital hepatic cysts have a good prognosis, and complications are rare<sup>[37]</sup>. The median follow-up of this study was 11.7 mo (range 1-36 mo). There were no reported postoperative complications or recurrences in this series of patients.

## CONCLUSION

Liver cysts in infants often pose significant diagnostic challenges, and paediatric surgeons should be aware of the characteristics of liver cysts to ensure appropriate treatment. Here, we report our experience with congenital hepatic cysts in infants and review the recent literature. Additional prospective and large-scale studies are warranted.

## **ACKNOWLEDGEMENTS**

The authors thank Mr. Xiurui Lv (Nanjing Children's Hospital Affiliated to Nanjing Medical University) for language polishing.

## **Figure Legends**

**figure 1** Ultrasound imaging of case 11 with a solitary intrahepatic biliary cyst. A: Ultrasound scan shows a large cyst in the abdomen (red arrow); B: Computed tomography scan shows two large intraabdominal cysts (red arrow).

**Table 1 Clinical presentation of patients with simple hepatic cysts**

Patient	Sex	Premature	Symptoms	USG	CT/MRI/UGI	Surgery	Complications
1	F	No	Jaundice	34 w prenatally	CT	L unroofing	None
2	M	No	Vomit	26 w prenatally	UGI	L unroofing	None
3	M	No	Enlargement	38 w prenatally	MRI	L-C unroofing	None
4	F	No	Confusing diagnosis	28 w prenatally	MRI	L CE	None
5	M	No	Abdominal distension	39 w prenatally	CT	CE	None
6	F	No	Enlargement	24 w prenatally	CT	L-C CE	None
7	M	No	Vomit, abdominal distension	26 w prenatally	MRI	L-C CE	None
8	M	No	Confusing diagnosis	/	/	Unroofing	Omphalocele
9	M	34 + 2 w	Enlargement	26 w prenatally	CT and MRI	L-C unroofing	Polycystic kidneys
10	M	33 + 4 w	Confusing diagnosis	/	/	CE	Omphalocele

F: Female; M: Male; USG: The first time of finding a cyst prenatally by ultrasonography; CT: Computed tomography; MRI: Magnetic resonance imaging; UGI: Upper gastrointestinal examination; L: Laparoscopy; L-C: Laparoscopy converted to open; CE: Cyst excision.

**Table 2 Histologic features of simple hepatic cysts**

Patient	Type of Cyst	Size (cm)	Epithelial lining	Cytokeratin	Fluid
1	Solitary	3 × 4 × 4	Cuboidal	Not available	Yellowish
2	Solitary	7 × 6 × 4	Cuboidal	Positive	Yellowish
3	Solitary	8 × 8 × 6	Cuboidal	Not available	Yellowish
4	Solitary	3 × 4	Cuboidal	Positive	Yellowish
5	Solitary	6 × 5 × 4	Cuboidal	Not available	Yellow
6	Solitary	4 × 3 × 2	Cuboidal	Positive	Yellow
7	Solitary	4 × 5	Cuboidal	Positive	Yellow
8	Solitary	2 × 2 × 1	Not Observed	Not available	Not described
9	Multiple	6 × 9 × 6 in right; 1.5 × 0.5 in left	Cuboidal	Positive	Yellowish
10	Multiple	Not described	Cuboidal	Positive	Not described

Size: The size of cysts during operation.

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