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Congenital solitary intrahepatic biliary cyst in infant: A rare case report

Anil Kumar Yadav [a,*], Jianghua Zhan [b], Shujian Zhang [b], Nita Yadav [a]

[a] The Graduate School of Tianjin Medical University, 300070, Tianjin, China
[b] Department of Pediatric Surgery, Tianjin Children's Hospital, Tianjin, 300134, China

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ABSTRACT

Solitary liver cysts (SLCs) are usually asymptomatic, uncommon and nonparasitic cysts of congenital origin, most frequently discovered during imaging studies. It may develop in the form of simple cyst and biliary cyst. The majority of the cysts that are discovered incidentally remain small. The etiology of SLC is unknown. Perioperative cholangiography is helpful to confirm the connection between cyst and biliary tree. In several clinical situations, including resection of malignant or benign biliary lesions, reconstruction of the biliary system using the Roux-en-Y jejunum limb should be performed as the standard procedure.

1. Introduction

Solitary liver cysts (SLCs) rarely manifest in infancy. They are unilocular, benign lesions of the liver and can be defined as a simple cyst or a biliary cyst according to the composition of the cyst fluid and the cyst's anatomical relationship with the biliary tree [1]. SLCs are usually asymptomatic [1]. Perioperative cholangiography should be performed in all cases to detect connections between the SLC and the biliary tree [5]. Management options include percutaneous aspiration, injection of sclerosing agents, laparoscopic or open fenestration, and surgical cystectomy or Roux-en-Y hepaticojejunostomy [2,5].

The aim of this study was to describe the clinical, imaging features, differential diagnosis, histopathological findings and the surgical options for cyst.

2. Case report

This full term male infant was antenatally diagnosed with cystic abdominal mass at 28 weeks of gestation. Prenatal diagnosis could not confirm the exact origin of cyst. The diagnostic options could be mesenteric cyst, duplication cyst, choledochal cyst and omental cyst. Postnatal abdominal ultrasound demonstrated cystic mass but the baby was asymptomatic. At 12 months of age, he was admitted to our hospital for further evaluation. In laboratory findings, total bilirubin 4.8micromol/l, direct bilirubin 2.4micromol/l, ALT 12U/L. PT, alkaline phosphate and albumin were within normal range (Table 1a). Routine urine examination, renal function test and chest x-ray were normal. A repeat abdominal ultrasound showed a 5.9 × 4.7 × 5.0 cm homogeneous anechoic hepatic hilum cystic mass. The computed tomography (CT) revealed the cystic mass in the right hepatic lobe, and the size is 5.4 × 4.8 cm, extending to the hepatic hilar region (Fig. 1a). Magnetic resonance imaging (MRI) showed an irregular single mass in the right hepatic lobe, which is about 5.1 × 4.6 × 4.3 cm, adjacent intrahepatic duct was slightly pressed, the mass extending to the hepatic hilar region and has close relationship with intrahepatic bile duct (Fig. 1b). Magnetic resonance(MR) scan was performed to further define the anatomy but it failed to confirm the connection between cyst and biliary tree. As the diagnostic dilemma continued, laparotomy was done. In the meantime echocardiography demonstrated the PDA, which was ligated and the Patient was readmitted after 8 months. We performed a right subcostal incision, which revealed a large cystic mass in the center of the right lobe of liver. The cyst wall was opened which were filled with fluid. Intraoperative cholangiography was done which confirmed the connection between the cyst and biliary

* Corresponding author. Tel.:+86 13752093515; fax: +86 22 23525331.
E-mail addresses: dranil2042@gmail.com (A.K. Yadav), zhanjianghuatj@163.com (J. Zhan).

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Table 1a
Liver function tests before operation.

Normal range	Results	Tests
0–40	12 U/L	ALT
60–320	167 U/L	ALP
8–40	36.4 U/L	AST
5.1–18.8	4.80 μMOL/L	TBil
1.7–6.8	2.40 μMOL/L	DBil
3.4–10.3	2.40 μMOL/L	IBil
130–280	287 U/L	LDH
35–55	43.60 g/l	Albumin

tree and biliary strictures (Fig. 1c). The cyst was located close to and has connections with the intrahepatic biliary ducts. Because of biliary strictures, cyst was completely excised and a Roux-en-Y hepaticojejunostomy was performed with enteric drainage (Fig. 2a) and liver biopsy was taken. Perioperative diagnosis was a solitary intrahepatic biliary cyst.

Postoperative period was uneventful. He was discharged on 14th post-operative day. Followup 3 monthly for 1 year, with abdominal ultrasound and liver function test (Table 1b). Histopathology showed the wall is composed of fibrous tissue, hemorrhagic cystic wall, focal necrosis, infiltration of inflammatory cells, few areas lined with simple cuboidal epithelium (Fig. 2b).

3. Discussion

Congenital non parasitic liver cysts are categorized as solitary or multiple; the latter is associated with polycystic kidney disease and Caroli's disease. Solitary liver cyst (SLC) never involves other organ [1,3], and is further classified as simple solitary liver cyst and solitary intrahepatic biliary cyst (SIBC). The etiology of SLC is unknown. However it has been hypothesized that obstruction of aberrant bile ducts from infection or as a result of a vascular disruption during the fetal period can be a cause [1,3,4]. But these etiologies does not hold true in our case. SLC is located to and connections with the intrahepatic bile ducts. It is likely that solitary liver cyst is of biliary duct origin.

SLCs are usually asymptomatic and their symptoms do not manifest until patients are in their forties or fifties [1]. The anteroinferior segment of the right hepatic lobe is the most frequent site of SLC [1,4].

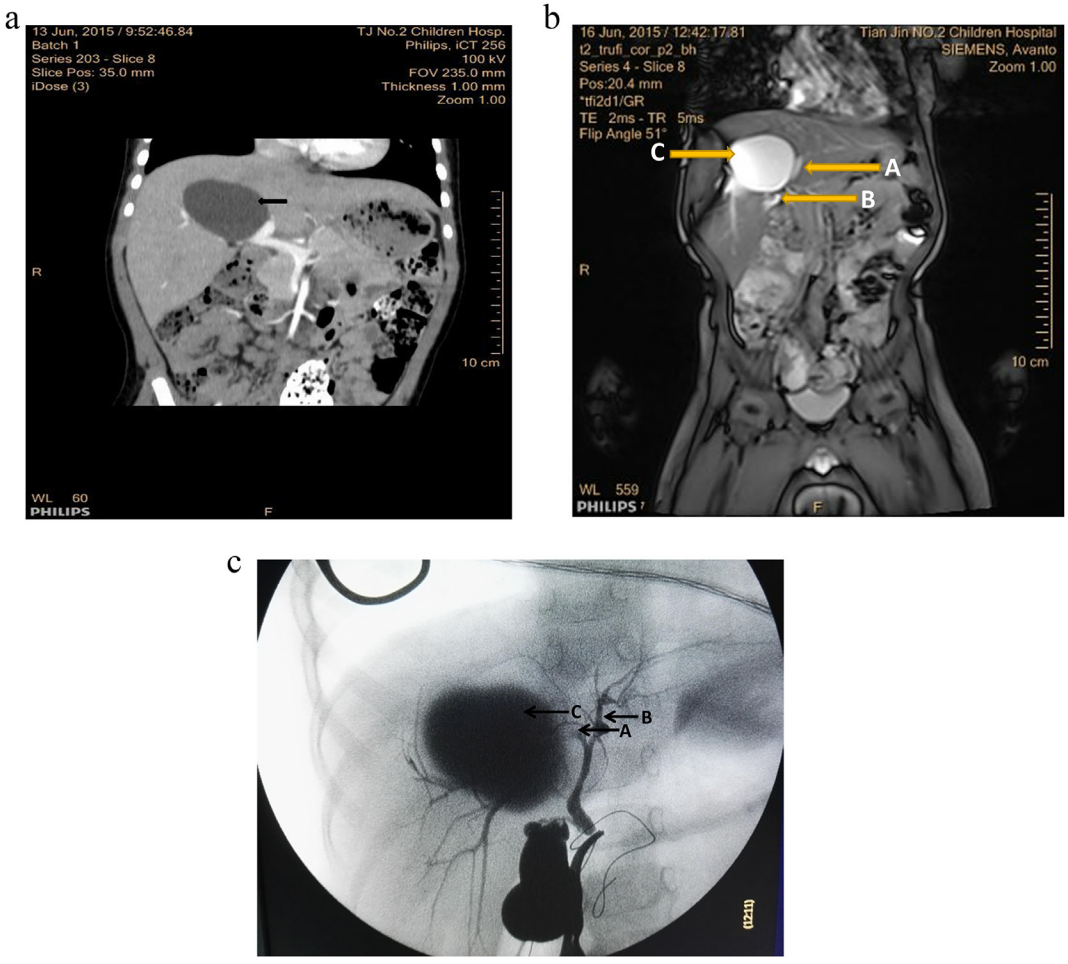

Fig. 1. a: CT scan (arrow) shows cystic mass in the right hepatic lobe, the mass boundary is irregular, the size is about 54.1 * 48.6 mm, extending to the hepatic hilar region. Fig. 1b: MRI showing an irregular mass in the right hepatic lobe, which is about 5.1 × 4.6 × 4.3 cm, the border is clear. Adjacent intrahepatic duct is lightly pressed, the mass extending to the hepatic hilar region and has close relationship with intrahepatic bile duct. (A. left hepatic duct, B. right hepatic duct, C.cyst). Fig. 1c. Intraoperative cholangiogram showing the cyst is communicated with intrahepatic bile duct. (A. right hepatic duct, B. left hepatic duct, C. cyst).