

## SEPTATE GALLBLADDER: A REPORT OF TWO CASES

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Congenital anomalies of the gallbladder are rare, and a wide range of anatomical malformations affecting its shape, size, number, and position have been described.<sup>1,2</sup> Septate gallbladder is among the malformations that has not been well documented, because it is usually asymptomatic or discovered accidentally during the evaluation of jaundice or at postmortem examination, but at times it may result in pitfalls at ultrasound imaging, causing a false-positive diagnosis of gallstones.<sup>3-5</sup> Rarely, however, septate gallbladder causes recurrent attacks of abdominal pain or become complicated by cholelithiasis.<sup>6,7</sup> This report describes two children, one of them with recurrent attacks of abdominal pain who was found to have a septate gallbladder with cholelithiasis on abdominal ultrasound, and the other with recurrent attacks of abdominal pain and jaundice who was confirmed to have a septate gallbladder by ERCP.

### Case Reports

#### Case No. 1

A four-year-old female presented to the hospital with recurrent attacks of abdominal pain of about a year's duration. The pain was colicky in nature and sometimes associated with vomiting. There was no history of sickle cell disease or any other blood disorder. Examination revealed a healthy young girl with no other abnormalities. Her CBC, liver function tests and electrolytes were normal. Abdominal ultrasound (Figure 1) revealed a septate gallbladder with normal wall thickness and multiple gallstones. She underwent laparoscopic cholecystectomy which was converted to open cholecystectomy due to problems with the gas flow. Her condition improved after the operation and was discharged home on the third postoperative day. Histology of the gallbladder showed a septate gallbladder with a septum near the fundus and multiple gallstones. The gallbladder also showed evidence of chronic cholecystitis.

#### Case No. 2

An 11-year-old female was admitted to the hospital with obstructive jaundice, itching, skin rash and history of abdominal pain of one month's duration. Examination revealed a thinly-built girl with generalized macropapular skin rash and generalized scratch marks. Abdominal examination was normal. Her investigations were as follows: total bilirubin =9.3 mg/dL, direct bilirubin =7.9 mg/dL, alkaline phosphatase =1108  $\mu$ mL, AST =88  $\mu$ mL, ALT =112  $\mu$ mL, total protein =8.4 g/dL, Hb =12.1 g/dL, Hct =36.1%, RBC =5.06x10<sup>12</sup>/l, WBC =6.8x10<sup>3</sup>/mm<sup>3</sup>, platelets =405, 000/mm<sup>3</sup>. Her hepatitis B and C were negative. Abdominal ultrasound and CT scan were normal. She was treated conservatively and her condition improved.

Eight months later she was admitted again with similar complaints. She was slightly jaundiced with total bilirubin =3.6 mg/dL, and direct bilirubin =1.72 mg/dL. Her hepatitis profile and autoantibodies were negative. Her AST =107  $\mu$ mL, ALP =869  $\mu$ mL, ALT =122  $\mu$ mL and total protein =8.6 g/dL. Her abdominal ultrasound this time revealed a contracted gallbladder with a thick wall. Adjacent to the gallbladder was another cystic swelling 18x16x13 mm in size. The diagnosis of choledochal cyst was suggested (Figure 2). ERCP showed normal bile ducts and septate gall bladder (Figure 3). The patient underwent open cholecystectomy and liver biopsy. Intraoperatively, she was found to have a septate gallbladder but no gallstones. Histology of the gallbladder showed a septate gallbladder with chronic cholecystitis. Her liver biopsy was suggestive of early sclerosing cholangitis. Postoperatively, she did well and was discharged home on the seventh post-operative day.

She was re-admitted again to the hospital five months later with jaundice and skin rash. Her investigations were as follows: total bilirubin = 3.6 mg/dL, direct bilirubin =2.4 mg/dL, AST =103  $\mu$ mL, LDH =211  $\mu$ mL, ALT =137  $\mu$ mL, GGT =32  $\mu$ mL, and amylase =39  $\mu$ mL. She was treated conservatively and was discharged home five days later. Now she is well and being followed up in the Gastroenterology clinic.

### Discussion

Congenital anomalies of the gallbladder have been classified into malformations of shape, number, site, size

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FIGURE 1. Abdominal ultrasound showing a septate gallbladder and cholelithiasis.

FIGURE 2. Abdominal ultrasound showing a septate gallbladder simulating a choledochal cyst

FIGURE 3. ERCP showing normal bile ducts and a septate gallbladder.

and heterotopias.<sup>1,2</sup> These anomalies may be asymptomatic discovered accidentally or may cause a wide range of complications. With recent advances in ultrasonography, it is now possible to diagnose these anomalies even prenatally and as early as the 14th week of gestation. In a prenatal study of 10,016 cases of fetal gallbladder, Bronshtein et al. found 17 (0.15%) cases of fetal anomalies, two of which were septate or bilobed gall bladder.<sup>8</sup>

Septate gallbladder is characterized by the presence of a septum that divides the gallbladder into two chambers. When the septum dividing the gallbladder lies longitudinally, it is called bilobed gallbladder, and when there is a transverse septum separating the fundus from the rest of the gallbladder, it is called an hour-glass gallbladder. Normally in an hour-glass gallbladder, the two compartments communicate through an opening in the septum that is variable in size.<sup>2,5,7,9</sup> Septate gallbladder most likely results from incomplete resolution of the solid stage of gallbladder development that is present before the third fetal month. These gallbladder septate are most commonly single, but multiseptate gallbladder, as well as postinflammatory adhesions and compartmentalization of the gallbladder have also been described.<sup>3</sup> In our patients, these septate were most likely congenital in origin. This is supported by the young age of our patients, and negative history of acute cholecystitis. The histological evidence of chronic cholecystitis is attributed to gallstones in our first patient, and possibly due to small stones that had passed spontaneously in the second one. Bile stasis and subclinical infection without stone formation is another contributing factor in our second patient.

In rare instances, a septate gallbladder may cause recurrent attacks of abdominal pain or become complicated by cholelithiasis. Our first patient suffered from repeated attacks of abdominal pain for one year. The young age of the patient, and rarity of cholelithiasis and cholecystitis at this age contributed to the delay in the diagnosis. The diagnosis was however established only after an abdominal ultrasound was ordered. In this patient, the primary disease could be cholelithiasis complicated by minor cholecystitis responsible for her symptoms, and the septate gallbladder could be a congenital nonsignificant, incidental finding. The possibility of bile stasis secondary to gallbladder septation as the predisposing factor for cholelithiasis could not however be excluded. Our second patient suffered from recurrent episodes of abdominal pain, skin rash and jaundice. Granot et al. reported a four-year-old girl with

duplication of the gallbladder associated with obstructive jaundice.<sup>10</sup> In our second patient, the recurrent attacks of jaundice were initially thought to be due to the ultrasonic misdiagnosis of choledochal cyst, but subsequently this proved to be due to sclerosing cholangitis. The primary disease in this patient might be chronic cholecystitis with no stones at diagnosis and the cholangitis, which was diagnosed by liver biopsy, support the possibility of transient obstruction caused by passing biliary stones. The septate gallbladder could be of developmental etiology with post inflammatory disease producing internal compartmentalization. The congenital origin of the septate gallbladder could not however be excluded.

We found ERCP valuable in establishing the diagnosis in our second patient. The fact that our patient was admitted to the hospital postoperatively with jaundice but without abdominal pain is indirect evidence that the septate gallbladder was a contributing factor to the abdominal pain, and although no gallstones were found at operation, the possibility that small stones had passed cannot be ruled out. In conclusion, gallbladder abnormalities, although rare should always be considered in the differential diagnosis of children presenting with recurrent attacks of abdominal pain, and abdominal ultrasound should form part of their investigations.

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