The use of laparoscopic subtotal cholecystectomy in a case with a cholecystohepatic duct

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A cholecystohepatic duct is a rare biliary anomaly that is identified by the drainage of the hepatic duct into the gallbladder [1,2]. The incidence of this anomaly is less than 1% [3,4]. We herein report on the case of a 2-year-old boy with a cholecystohepatic duct that originated from the right posterior segment of the liver and flowed into the neck of the gallbladder. We performed a laparoscopic subtotal cholecystectomy on this patient. This is the first case report of a cholecystohepatic duct treated using a laparoscopic subtotal cholecystectomy.

1. Case report

A 2-year-old boy was admitted with jaundice and white stool. The child had a supralevator anorectal malformation with a rectourethral fistula, scrotal hypospadias and vesicoureteral reflux. Previously, he had undergone a correction of the anorectal malformation, and had a Cohen ureteric reimplantation to treat the vesicoureteral reflux. An ultrasound examination and a computed tomography (CT) scan had revealed gallstones one year prior to being admitted, and he had been followed without therapy due to the lack of symptoms.

The patient was in good condition. Palpation of the abdomen revealed no abnormalities. His total bilirubin level was 2.7 mg/dl with a direct fraction of 1.9 mg/dl. His alkaline phosphatase (ALP) level was measured at 3015 IU/L, his γ-glutamyl transpeptidase (γ-GTP) level was 597 IU/L, and his aspartate aminotransferase level was 247 IU/L. The number of leukocytes and the levels of C-reactive protein (CRP) were within normal limits.

An ultrasound examination and magnetic resonance cholangiopancreatography (MRCP) showed gallstones, a dilated gallbladder, and dilated intrahepatic bile ducts. However, there was no dilation identified in the common bile duct (Fig. 1).

The patient underwent a percutaneous transhepatic gallbladder drainage procedure (PTGBD) that relieved the jaundice. Cholangiogram performed via the PTGBD tube revealed that the right posterior segmental branch flowed into the neck of the gallbladder, and the right anterior segmental branch entered into the left intrahepatic duct (Fig. 2). The diagnosis of a cholecystohepatic duct was thus established, and a laparoscopic subtotal cholecystectomy was performed. The gallbladder was dissected meticulously from the fundus downward. The PTGBD tube was removed during the dissection. A cord structure, which was considered a cholecystohepatic duct, was noted between the liver and the gallbladder (Fig. 3). We stopped the dissection when the cord structure...
was noted, and an intraoperative cholangiogram showed the cholecystohepatic duct flowed into the neck of the gallbladder and a preserved cystic duct. Therefore, the gallbladder was subtotally resected (almost three-fourth of the gallbladder was resected), thus leaving the cholecystohepatic duct and cystic duct intact, and the residual gallbladder was sutured laparoscopically (Fig. 4). One day after the operation, the leakage of bile developed from the drain. The bile leakage was managed conservatively, and had disappeared completely by postoperative day three. The patient was discharged fifteen days after the operation. Through six months of follow-up, the patient has been healthy without any recurrence of biliary stones or biliary stasis.

2. Discussion

A cholecystohepatic duct is a rare biliary anomaly characterized by the drainage of the extrahepatic duct directly into the gallbladder [1,2]. Benson et al. reported that the incidence of a cholecystohepatic duct was found to be 0.7% in a series of cholecystectomies [3]. Champetier et al. reported that the combined incidence of a cystohepatic duct (characterized by the drainage of the extrahepatic duct directly into the cystic duct) and a cholecystohepatic duct was 0.85% [4]. The following types of cholecystohepatic ducts have been reported [2,5]: the common hepatic duct draining into the gallbladder, the right and left hepatic duct draining separately into the gallbladder, the right hepatic duct draining into the gallbladder [6–9], and the aberrant hepatic duct draining into the gallbladder.

Embryologically, the hepatic diverticulum arises ventrally from the endodermal lining of the foregut during the fourth week.
gestation [2,4,10]. The hepatic diverticulum is divided in two, between the pars hepatica and the pars cystica. The pars hepatica represents the anlage of the liver, the intrahepatic duct, and the common hepatic duct. The pars cystica differentiates into the gallbladder, the cystic duct, and the common bile duct. It is believed that a delay in the separation of the pars hepatica and the pars cystica can lead to the formation of a cholecystohepatic duct.

To our knowledge, more than thirty cases of cholecystohepatic duct have been reported, but the incidence of the anomaly with drainage of the right hepatic duct directly into the gallbladder is extremely low. Only five such cases have so far been reported [6–9]. In three of these cases, a subtotal cholecystectomy was performed [6,7]. In one of these cases, a percutaneous transhepatic cholangiography drainage (PTCD) procedure and a cholangioscopic lithotripsy were performed [8]. In the final case, a cholecystectomy and a hepaticojejunostomy were performed [9]. In all five cases, the postoperative courses were uneventful.

In the pediatric age group, seven cases of cholecystohepatic ducts were reported, including the present case [8,11–14] (Table 1). Interestingly, five cases also had other malformations. Redkar et al. reported on two cases of cholecystohepatic duct associated with esophageal atresia [14]. They suggested that a common defect during the mesodermal development could explain both the biliary abnormality and the association with esophageal atresia.

The preoperative recognition of a cholecystohepatic duct is very important. If the cholecystohepatic duct only drains a hepatic territory to a limited extent, the iatrogenic injury of the cholecystohepatic duct during surgical intervention will cause serious complications such as jaundice, liver atrophy, or liver dysfunction. Especially in pediatric cases, computer tomography (CT) scans and magnetic resonance cholangiopancreatography (MRCP) are not necessarily helpful as a diagnostic tool in the identification of a biliary anomaly due to the size of the patient. In the present case, MRCP revealed a dilated gallbladder, dilated intrahepatic bile ducts, and gallstones, but no cholecystohepatic duct was observed prior to performing cholangiogram via the PTGBD tube. In five of eight pediatric cases, preoperative cholangiogram was performed (Table 1). It is considered that preoperative cholangiogram is necessary for cases of cholecystohepatic duct, especially in pediatric cases, because of the difficulty of obtaining accurate images of a cholecystohepatic duct on a CT scan or by MRCP. In the present case, a laparoscopic subtotal cholecystectomy was performed. This case study is the first report of the treatment for a cholecystohepatic duct. The use of laparoscopy provided a better opportunity for observation which resulted in the safe dissection of the gallbladder without a cholecystohepatic duct injury.

Obstructive jaundice is the clinical presentation in patients with a cholecystohepatic duct. The insufficient drainage from the gallbladder via the cystic duct causes the presentation of jaundice. In the present case, the common hepatic duct was considered to be heavily opposed by the dilated gallbladder, which led to jaundice. There are two reasons why subtotal cholecystectomy was performed on this patient, rather than cholecystectomy and hepaticojejunostomy. First, it was supposed that subtotal cholecystectomy would lower the risk of the re-expansion of the gallbladder. Second, the diameter of the cholecystohepatic duct, which was less than 3-mm, was estimated to be too small to perform a hepaticojejunostomy without risking a postoperative anastomotic stricture. At the present time, the patient has been healthy without any recurrence of biliary stones or biliary stasis. Therefore, a subtotal cholecystectomy is considered to be a useful procedure for patients with a cholecystohepatic duct.

Acknowledgments

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References


Table 1

<table>
<thead>
<tr>
<th>Author</th>
<th>Age &amp; gender</th>
<th>Associated anomaly</th>
<th>Type of the bile duct that drains into the gallbladder</th>
<th>Perioperative cholangiogram</th>
<th>Operative procedure</th>
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<td>Stringer</td>
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<td>Right and left hepatic duct</td>
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<td>Cholecystectomy + hepaticojejunostomy</td>
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<td>Muramatsu</td>
<td>7 years old (F)</td>
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<td>Common hepatic duct</td>
<td>PTC</td>
<td>Subtotal cholecystectomy + hepaticocholedochoenterostomy</td>
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<td>Redkar</td>
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<td>ERC</td>
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<td>Redkar</td>
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<td>Iwasaki</td>
<td>3 years old (F)</td>
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<td>Right posterior segmental branch</td>
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<td>Kawakubo (present study)</td>
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<td>Right posterior segmental branch</td>
<td>PTGBD</td>
<td>Laparoscopic subtotal cholecystectomy</td>
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