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Unsuspected Duplicated Gallbladder in a Patient Presenting with Acute Cholecystitis

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INTRODUCTION

Duplicated gallbladder (GB) is a rare congenital anomaly that is found in 0.026% of autopsies (1). Although the disease is rare, preoperative diagnosis has some clinical implications. Undiagnosed duplicated GB may lead to cholecystectomy, leaving another GB or injury to adjacent structures (2). Preoperative diagnosis of duplicated GB enables the surgeon to evaluate its type before selecting the most appropriate surgical technique (3). We describe a patient with acute cholecystitis caused by duplicated GB, which was evaluated preoperatively and treated laparoscopically.

CASE DESCRIPTION

An 80-year-old man presented at an emergency department with right upper quadrant abdominal pain lasting for 3 days. The patient was healthy without any underlying disease. Physical examination revealed right upper quadrant tenderness. Laboratory tests revealed elevated C-reactive protein (48.3 mg/dL), bilirubin (1.71 mg/dL), aspartate aminotransferase (310 IU/L), and alanine transaminase (204 IU/L) concentrations. Abdominal computed tomography (CT) showed thickened GB wall with pericholecystic fluid collection, consistent with acute cholecys-

Duplicated gallbladder (GB) is a rare congenital disease. Surgical management of a duplicated GB needs special care because of concurrent bile duct anomalies and the risk of injuring adjacent arteries during surgery. An 80-year-old man visited an emergency room with right upper quadrant abdominal pain. Computed tomography (CT) revealed cholecystitis with a 2-bodied GB. Because of this unusual finding, magnetic resonance choledochopancreatography was performed to detect possible biliary anomalies. The 2 GB bodies were unified at the neck with a common cystic duct, a so-called V-shaped duplicated GB. The patient's right posterior hepatic duct joined the common bile duct (CBD) near the cystic duct. The patient underwent laparoscopic cholecystectomy without adjacent organ injury, and was discharged uneventfully. Surgeons should carefully evaluate the patient preoperatively and select adequate surgical procedures in patients with suspected duplicated GB because of the risk of concurrent biliary anomalies.

Keywords: Duplicated Gallbladder; Cholecystitis; Cholecystectomy; Laparoscopy

titis. Furthermore, a 2-body GB with an impacted GB stone in the common cystic duct was observed without common bile duct (CBD) stones. Magnetic resonance cholangiopancreatography (MRCP) was performed to assess the biliary anomalies. MRCP also showed a duplicated GB, in which one GB (GB 1) was inserted into the neck of the other GB (GB 2), with a common cystic duct. The right posterior bile duct joined the CBD near the common cystic duct. The patient was diagnosed with acute cholecystitis caused by duplicated GB (Fig. 1). Therefore, we performed laparoscopic cholecystectomy on 5 October 2015.

After induction of general anesthesia, 3 ports were inserted as usual. After dissection around Calot's triangle, a common cystic duct with 2 GB bodies was found. The right posterior bile duct was identified above the cystic duct. After milking the cystic duct using laparoscopic Maryland dissecting forceps owing to the impacted GB stone detected on preoperative CT, the cystic duct was ligated using Hem-o-lock[®] clips (WECK Closure System; Teleflex Inc., Morrisville, NC, USA). An abdominal drain was placed under the liver and the GB was retrieved via the umbilical port in a protective bag. The abdominal drain was removed on postoperative day 1 after checking for bile leakage or bleeding. The patient was discharged uneventfully on postoperative day 3.

The pathologic results showed separation of the lamina propria and smooth muscle, and the perimuscular connective tis-



Fig. 1. Magnetic resonance choledochopancreatography reveals V-shpaed duplicated GB with right hepatic duct anomaly. (A) GB 1 was inserted into the neck of the other GB (GB 2). (B) The right posterior bile duct (arrow) joined the CBD near the common cystic duct. GB = gallbladder, CBD = common bile duct.



Fig. 2. Macroscopic view (A) of the specimen and microscopic features of the duplicated GB (B). The GB body shows separation of the lamina propria and muscle layer (arrowhead). Perimuscular connective tissue (arrow) is shared between the GB 2 bodies with H & E staining (\times 40). GB = gallbladder, H & E = hematoxylin and eosin.

sue was fused between the 2 GBs (Fig. 2). The findings were consistent with the typical microscopic findings of duplicated GB.

DISCUSSION

Duplicated GB was first reported by Blasius in 1675, and was classified by Boyden in 1926 based on embryological differences between the types of duplicated GB. In 1975, Harlaftis et al. (1) modified the classification into 2 types, which focused on the most commonly detected types. The GB originates from the cystic primordium. Type 1 duplicated GB originates from a single cystic primordium and it splits during embryogenesis approximately 5–6 weeks after fertilization. Irregular growth of the split cystic primordium yields 2 GB bodies. Earlier bifurcation results in more complete duplication. Fig. 3 shows representative images of type 1 duplicated GBs, which include the septat-



Fig. 3. Classification of duplicated GB. Adapted from Harlaftis et al. (1). GB = gallbladder.

ed type (10.8%), V-shaped type (9.5%), and Y-shaped type (24.3%). The septated GB has a longitudinal septum that extends from the fundus to the neck of the GB. The V-shaped GB comprises 2 GB bodies, which are joined at the neck via a common cystic duct. The Y-shaped GB comprises 2 complete GBs together with 2 cystic ducts, which join at the common cystic duct before entering the CBD.

Type 2 duplicated GBs originate from multiple cystic primordia to form multiple GBs and cystic ducts, which enter the CBD independently. In some cases, the cystic duct enters the left or right hepatic duct. The most common type 2 duplicated GBs are the ductular type (48.6%) and the trabecular type (2.7%). In ductular duplicated GBs, 2 independent cystic ducts join the CBD independently. Trabecular duplicated GBs have peculiar anatomies. The accessory cystic duct enters the right or left hepatic duct (1,4).

Although high-resolution ultrasonography is useful for the diagnosis of duplicated GB, it is often unable to delineate the anatomic relationship, which is essential to confirm the anatomical type (5). Endoscopic retrograde cholangiopancreatog-raphy (ERCP) provide accurate visualization of the biliary anatomy, including duplicated GBs. However, it should be reserved for managing concurrent CBD stones or cholangitis because of its invasiveness. MRCP is a relatively non-invasive modality compared with ERCP, and provides definitive images, including 3 dimensional (3D) reconstructed images that reveal the anatomical relationships between adjacent structures (6). Diagnostic imaging modalities enable differentiation of duplicated GB from

choledochal cyst, folded GB, GB diverticulum, or focal adenomyomatosis (7-9).

The management of duplicated GB is same as that of cholecystectomy of a single GB, but the surgeon needs to consider several things before surgery. In particular, preoperative awareness of duplicated GB is important. In several case reports, patients who underwent cholecystectomy owing to abdominal pain were diagnosed with remnant GB during surgery, and a second cholecystectomy was necessary (10). It is important to confirm the type of duplicated GB because of the clinical implications and embryological differences between each type (Fig. 3). Type 2 duplicated GB has independent cystic ducts, which enter the right or left bile duct. Because it is possible to injure the CBD or right hepatic artery during dissection, surgeons have recommended open surgery for careful dissection and adequate management of injured adjacent structures (11). However, laparoscopic surgery was reported to be feasible for type 1 duplicated GB because of the common cystic duct (12,13). Our case had a type 1 V-shaped duplicated GB, but the right posterior bile duct joined the CBD near the common cystic duct. Nonetheless, we performed laparoscopic cholecystectomy because the patient's general condition was good and his cholecystitis was not severe based on preoperative imaging. The common cystic duct was identified in the operative field, and was ligated without injuring the right posterior bile duct.

Although duplicated GB is very rare, its preoperative detection allows the surgeon to avoid injuring adjacent structures during surgery or repeated surgery. Patients with suspected duplicated GB should undergo preoperative MRCP or ERCP. Imaging modalities are useful to confirm the type of duplicated GB and select the most appropriate surgical approach.

DISCLOSURE

The authors have no potential conflicts of interest to disclose.

AUTHOR CONTRIBUTION

Conceptualization: Jeong CY. Data curation: Lee JK, Park JH, Kwag SJ. Writing - original draft: Lee W, Song DH, Kim JY, Lee YJ. Writing - review & editing: Lee W, Song DH, Kim JY, Lee YJ. Validation: Ju YT, Jung EJ, Hong SC, Choi SK.

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