Case Report

A Case of Obstructive Cholangitis of the Accessory Hepatic Duct Occurring 30 years after a Cholecystectomy

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Abstract

We reported a case which developed obstructive cholangitis of the accessory bile duct 30 years after undergoing a cholecystectomy. A 78-year-old male patient was admitted to the emergency department, presenting with a fever of 40 degrees Celsius with chills and abdominal pain. The patient’s past medical history included a cholecystectomy performed approximately 30 years ago.

Contrast-enhanced computed tomography (CT) and magnetic resonance cholangio-pancreatography (MRCP) findings led to a diagnosis of obstructive cholangitis associated with jaundice due to the obstruction of the accessory bile duct. This was possibly attributable to a ligation of the accessory bile duct which was injured during cholecystectomy. The intraoperative findings showed confluence of the dilated intrahepatic bile duct and the common bile duct in the vicinity of the cystic duct which was already resected. The accessory bile duct was ligated and resected at the confluence followed by right hepatic lobectomy. A histopathological examination of the resected specimen showed the cord-like, occluded area of the stenosed accessory bile duct with no neoplastic lesions. It should be noted that inflammation may occur in the accessory bile duct associated with difficult differentiation if cholecystitis develops concurrently due to gallstone incarceration in the neck of the gallbladder. Therefore, drip infusion cholangiography-CT and MRCP are required preoperatively for the sufficient visualization of direction of the bile ducts, and endoscopic retrograde cholangiography should also be performed if no satisfactory results are obtained from these imaging tests. It is important to ensure that the direction of the bile ducts in the individual segments is always confirmed before performing a cholecystectomy.

Key Words: accessory hepatic duct, bile duct injury, cholecystectomy

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Introduction

Embryologic anomalies of the bile ducts are common among those seen in the biliary tract\(^1\). In patients with this condition, hepatobiliary surgical procedures may be associated with intraoperative bile duct injuries if the operator is unaware of the presence of the condition. We encountered a patient who developed obstructive cholangitis of the accessory bile duct 30 years after undergoing a cholecystectomy. This report describes this case with some discussion.

Case Report

A 78-year-old male patient was admitted to the emergency department of this hospital in December 2007, presenting with a fever of 40 degrees Celsius with chills and abdominal pain. The patient’s past medical history included a low anterior resection of the rectum for rectal cancer and a cholecystectomy for acute cholecystitis. These procedures had been performed outside this hospital at 37 and 47 years of age, respectively. On admission, the patient had a height of 166 cm and body weight of 60 kg. The body temperature was 38.3 degrees Celsius with a pulse rate of 80 beats/min and blood pressure of 100/60 mmHg. No anemia but mild conjunctival icterus was noted. Palpation revealed tenderness in the upper abdomen which was flat and soft; however, no rebound tenderness or muscular guarding was observed.

Laboratory results showed markedly decreased white blood cell and platelet counts, thus suggesting sepsis due to severe infection. The serum AST, ALT, and total bilirubin levels were also slightly increased whereas the renal function was mildly decreased. The serum CA19-9 level, a tumor marker of the bile duct, increased to 140 U/mL (Table 1). Contrast-enhanced abdominal computed tomography (CT) showed significantly dilated intrahepatic bile ducts in the area of hepatic segments 5 and 6 with an atrophied parenchyma of the liver. There was an intrahepatic confluence of the dilated intrahepatic bile ducts and the common bile duct with a possible occlusion.
A case of Cholangitis of Accessory Hepatic Duct

near this region (Fig. 1). The wall of the occluded bile duct was slightly calcified with no irregularly enhanced region, and no neoplastic lesions could be observed. A diagnosis of obstructive cholangitis in the hepatic segments 5 and 6 was thus made, and percutaneous transhepatic biliary drainage (PTBD) was performed. Purulent bile was present in the dilated intrahepatic bile duct, and a bacterial culture of the bile showed *Escherichia coli*. The cholangitis rapidly improved after biliary drainage with no subsequent discharge of normal bile, and fluid was extremely watery. A cytological examination of the bile was performed in triplicate and the results were assessed to be Class II. The serum CA19-9 level which had been high on admission returned to the normal range as the cholangitis improved. Cholangiography using the PTBD tube revealed marked dilation of the intrahepatic bile duct with complete U-shape occlusion. However, no filling defect suggestive of gallstones was noted in the dilated bile duct (Fig. 2). Magnetic resonance cholangiopancreatography (MRCP) demonstrated U-shape occlusion of the dilated bile duct centrad, with no areas of signal void indicating either a gallstone or a neoplastic lesion. The occlusion was located below the bifurcation of the left and right hepatic ducts (Fig. 3a). On the other hand, drip infusion cholangiography (DIC)-CT showed no dilation of the bile duct but part of the bilateral hepatic ducts as well as the common bile duct (B7, B8).

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<th>Table 1 Laboratory data on admission</th>
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<tr>
<td>WBC 2,000/μl</td>
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<td>Hb 16.0 g/dl</td>
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<td>Plt 5.3 x10^9/μl</td>
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<tr>
<td>TP 6.2 g/dl</td>
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<td>Alb 3.6 g/dl</td>
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<td>T-Bil 3.1 mg/dl</td>
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<td>GPT 86 IU/l</td>
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<td>LDH 265 IU/l</td>
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<td>ALP 423 IU/l</td>
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Fig. 1 Contrast-enhanced abdominal CT scan shows significantly dilated hepatic ducts in the area of hepatic segments 5 and 6 with an atrophied parenchyma of the liver (arrow).

Fig. 2 Cholangiography using PTBD tube shows marked dilation of the intrahepatic bile duct with complete U-shape occlusion (arrow).

Fig. 3 MR-cholangiography demonstrates U-shape occlusion of the dilated bile duct (arrow) and the occlusion is located below the bifurcation of the left and right hepatic ducts (a). DIC-CT shows that the remnant of the cystic duct after cholecystectomy is also seen and located adjacent to the occluded area of the dilated bile duct (arrow) (b).
The remnant of the cystic duct after cholecystectomy was also seen and located adjacent to the occluded area of the dilated bile duct detected on MRCP and cholangiography (Fig. 3b).

These findings led to a diagnosis of obstructive cholangitis associated with jaundice due to the obstruction of the accessory bile duct. This was possibly attributable to a ligation of the accessory bile duct which was injured during cholecystectomy performed approximately 30 years ago. As the parenchyma of the liver at the occluded area atrophied, hepatic lobectomy including the dilated bile duct was needed and performed. The intraoperative findings showed confluence of the dilated intrahepatic bile duct and the common bile duct in the vicinity of the cystic duct which was already resected (Fig. 4a). There was significant atrophy of the hepatic parenchyma mainly at the segments 5 and 6 with compensatory hypertrophy of the left lobe. The accessory bile duct was thus ligated and resected at the confluence followed by right hepatic lobectomy. A resected specimen showed the cord-like, occluded area of the stenosed accessory bile duct with no neoplastic lesions. In the region supplied by the accessory bile duct, the parenchyma of the liver was observed to have markedly atrophied (Fig. 4b). Histopathologically, the occluded area was highly fibrotic and thick in association with infiltration of inflammatory cells; however, no malignant findings were observed in that area (Fig. 5). The patient’s postoperative course was uneventful, and the patient was discharged on the 16th hospital day. Nine months after surgery, the patient is now being treated on an outpatient basis with a favorable condition.

**Discussion**

The embryological origin of the intrahepatic bile duct, extrahepatic bile duct, and gallbladder is pars hepatica, pars cystica, and pars antrum, respectively\(^3\). Any anomalies of the bile duct may occur during such embryological development if there is a development
error/aberration or embryonic remnant of communication. The incidence of anomalies in the biliary tree is relatively common (3.1%), and among them, those in the cystic duct and the accessory bile ducts account for 2.1% and 1%, respectively. Different types of anomalies have also been reported regarding the confluence of the major bile ducts. The most common type, extrahepatic confluence of the left hepatic duct and right hepatic duct resulting from the right anterior branch joining with the right posterior branch, accounts for only 70% of all types of confluence, whereas the remaining 30% show different forms from each other.

In 1993, a group of Japanese researchers in the hepatobiliary field developed a classification system for such various anomalies of the biliary tract with multiple subtypes and malformations. According to the classification, anomalies of the bile ducts are divided into those in the aberrant bile duct or in the accessory bile duct, each of which is then subtyped. The accessory bile duct has also been referred to as aberrant hepatic duct; however, these terms are confusing because of lack of clear definition. Currently, the term of accessory bile duct is commonly used in light of the clinical and cholangiographic findings. The accessory bile duct can often be ligated and resected with minimal adverse effects if it measures around 2 mm in diameter, whereas it is generally necessary to reconstruct the biliary tract after a resection of the relatively large duct, which is the only route for bile excretion in the relevant hepatic segment.

The surgeon should therefore be careful not to overlook the accessory bile duct, which has been classified into types 0 to V by Hisatsugu et al., according to the positional relationship between the cystic duct and the confluence of the bile ducts. In cases of types 0, I, and V with the accessory bile duct joining into the cystic duct or the confluence of the common bile duct, common hepatic duct, and the cystic duct, the bile duct is likely to be injured during a cholecystectomy. In particular, it should be noted that inflammation may occur in the accessory bile duct associated with difficult differentiation if cholecystitis develops concurrently due to gallstone incarceration in the neck of the gallbladder. Therefore, DIC-CT and MRCP are required preoperatively for the sufficient visualization of direction of the bile ducts, and endoscopic retrograde cholangiography should also be performed if no satisfactory results are obtained from these imaging tests. Studies have reported intraoperative cholangiography (IOC) to also be useful for detecting the accessory bile duct, and it is meaningful to perform such an examination if it helps to prevent any injuries by visualizing clear images of the bile ducts. In addition, injuries of the bile ducts may be prevented by intraoperative procedures with a sufficient detachment of the cystic duct followed by its resection at the site as close as possible to the gallbladder.

In the present case, it was not feasible to classify the accessory bile duct according to the above system as approximately 30 years have passed since the cholecystectomy had been performed. However, retrospective evaluation indicated a possibility that this was a rare case of either a type I or V accessory bile duct, which might have been ligated during a cholecystectomy without identifying it as the accessory bile duct, and thereafter remaining occluded for a long period of time. The ligation of the accessory bile duct might therefore hinder or even abolish the ability of biliary excretion, thus leading to the onset of obstructive cholangitis with marked atrophy of the hepatic parenchyma in the supplied region. Therefore, a right hepatic lobectomy, including a resection of the atrophied area of the liver, was performed in the present patient, and this surgical modality is therefore considered to be appropriate for such a case because it also helped to control cholangitis.

Future advances in imaging diagnosis modalities such as computed tomography and magnetic resonance imaging are expected to make the differentiation of the accessory bile duct easier; however, it is important to ensure that the direction of the bile ducts in the individual segments is always confirmed before performing a cholecystectomy. Surgeons should also be familiar with types and incidence of anomalies of the bile ducts in order to prevent any injuries of the ducts during biliary tract surgery.

References