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Abstract
This report describes a rare case of hilar cholangiocarcinoma with intratumoral calcification that mimicked hepatolithiasis. A 73-year-old man presented to a local hospital with a calcified lesion in the hepatic hilum. At first, hepatolithiasis was diagnosed, and he underwent endoscopic stone extraction via the transpapillary route. This treatment strategy failed due to biliary stricture. He was referred to our hospital, and further examination suggested the existence of cholangiocarcinoma. He underwent left hepatectomy with caudate lobectomy and extrahepatic bile duct resection. Pathological examination revealed hilar cholangiocarcinoma with intratumoral calcification, while no stones were found. To the best of our knowledge, only one case of calcified hilar cholangiocarcinoma has been previously reported in the literature. Here, we report a rare case of calcified hilar cholangiocarcinoma and reveal its clinicopathologic features.

Key words: Cholangiocarcinoma; Klatskin tumor; Calcification; Lithiasis; Differentiation

Core tip: Our report describes an extremely rare case of hilar cholangiocarcinoma with intratumoral calcification. Imaging findings of this case were confusingly similar to those of hepatolithiasis, and it was extremely difficult to make an accurate diagnosis using available radiological findings. We performed pathological examination and observed hilar cholangiocarcinoma with intratumoral calcification. We herein report a rare case of calcified hilar cholangiocarcinoma and reveal its clinicopathologic features.


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INTRODUCTION
Calcification in the hepatic hilum is commonly caused by hepatolithiasis, which is defined as the presence of bile stones in the bile ducts proximal to the confluence of the right and left hepatic ducts\(^1\). The incidence of hepatolithiasis is as high as 18%-45% of patients with gallstone disease in East Asia\(^1\), and the disease has become more prevalent in Western countries due to an increase in migrants from endemic regions\(^2,3\). By contrast, hilar cholangiocarcinoma with intratumoral calcification is an extremely rare cause of calcification in the hepatic hilus, with only one previously reported case in the English literature\(^4\).

Although imaging findings are quite similar, the differential diagnosis between hepatolithiasis and calcified Klatskin tumor is crucial because of their different prognoses. We present a rare case of calcified hilar cholangiocarcinoma mimicking hepatolithiasis.

CASE REPORT
A 73-year-old man was admitted to a local hospital for investigation of a calcified lesion in the hepatic hilum associated with a dilated left intrahepatic bile duct. He initially received a diagnosis of hepatolithiasis. Endoscopic stone extraction via the trans-papillary route was attempted but was unsuccessful due to stricture of the left hepatic duct. He was referred to our hospital for further examination.

On admission, a blood test revealed mild elevation of carbohydrate antigen 19-9 (46.3 U/mL), though no elevation of carcinoembryonic antigen. All other laboratory findings, including serum concentrations of total bilirubin, aspartate aminotransferase, alanine aminotransferase, alkaline phosphatase and \(\gamma\)-glutamyl transpeptidase, were within normal ranges. Hepatic ultrasonography showed a 33.5 mm \(\times\) 26.5 mm intraductal mass at the confluence of the right and left hepatic duct. In addition, a highly echogenic mass with posterior acoustic shadowing was observed, suggesting a calcified lesion (Figure 1). Enhanced multi-detector-row computed tomography (CT) revealed marked dilatation of the left intrahepatic bile ducts upstream from the calcification and enhancement of the bile duct wall distal to the calcification (Figure 2). Endoscopic retrograde cholangiography (ERC) showed significant biliary stricture and disruption at the left hepatic duct, but no defect area (Figure 3). Furthermore, biopsy of the left hepatic duct was suggestive of adenocarcinoma.

Based on these findings, we diagnosed hilar cholangiocarcinoma complicated by the hepatolithiasis and decided to perform left hepatectomy with caudate lobectomy and extrahepatic bile duct resection. The patient underwent the scheduled operation. The operative time was 625 min, and the total blood loss was 1165 mL. No blood transfusion was required.

On histopathological examination, the tumor was found to have spread from the hepatic hilum to the left hepatic duct. The tumor was mucus-secreting...
and very hard, measuring 28 mm × 21 mm. It was a nodular-infiltrating type cholangiocarcinoma mainly existing in the perihilar bile duct with an abundance of fine calcification. No stones were found by the gross examination in the hilar lesion (Figure 4).

A regional lymph node in the hepatoduodenal ligament was invaded by tumor cells. Microscopically, this tumor was a gastric foveolar type adenocarcinoma and was rich in mucus. On the other hand, the majority of the calcified material was located within or replaced the tumor glands (Figure 5).

The postoperative course was uneventful, and the patient was discharged 21 d after surgery. At 7 mo after the surgery, the patient was well, without evidence of a recurrence of the disease.

DISCUSSION

This case report represents an unusual calcified hilar cholangiocarcinoma, which mimicked hepatolithiasis in clinical findings. To the best of our knowledge, there is only one article describing calcified Klatskin tumor in the literature⁴.

The typical cause of calcified lesion in the porta hepatis is a calculus; in patients with primary calcium bilirubinate hepatolithiasis, stones are frequently located in the large bile ducts, such as the main hepatic ducts⁵. Histologically, in patients with hepatolithiasis, fibrotic changes in the bile duct walls and periductal hepatic parenchyma are often seen and lead to stricture formation⁶,⁷. Furthermore, hepatolithiasis is closely associated with concomitant cholangiocarcinoma with an incidence of approximately 5%⁸, and this malignant lesion is also associated with biliary stricture⁹. Thus, among patients with hepatolithiasis, a calcified lesion is often associated with a biliary stenosis lesion caused by periductal fibrosis or cholangiocarcinoma⁹.

In the present case, initial imaging findings showed...
In conclusion, we presented an extremely rare case of hilar cholangiocarcinoma with intratumoral calcification. Radiological findings of this case were similar to those of hepatolithiasis. The pathogenic mechanism of this calcification was thought to be a dystrophic process that was associated with mucus.


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