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Benign Left Intrahepatic Biliary Stricture: A Case Report

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Abstract | **Case Report**

A biliary stricture is defined as a narrowing of the extrahepatic or intrahepatic biliary system leading to impaired antegrade bile flow, upstream biliary dilatation, and pathological consequences of biliary obstruction. Biliary strictures represent a significant diagnostic and therapeutic challenge. A multidisciplinary approach is recommended to establish the diagnosis and guide management, integrating biochemical results, imaging findings, and histopathological assessment. Malignancy is the most common etiology, highlighting the need for a high index of suspicion during evaluation. We report the case of a 64-year-old man who presented with febrile jaundice and right upper quadrant pain. Magnetic resonance imaging revealed a benign-appearing stricture of the left hepatic duct associated with dilatation and intrahepatic lithiasis of the left biliary ducts. A left hepatectomy was performed, and histopathological examination confirmed a benign biliary stricture. The aim of this report is to describe the diagnostic and therapeutic management of left-sided biliary strictures and to emphasize that, despite the high frequency of malignant causes, benign etiologies should never be excluded a priori.

Keywords: Cholangiocarcinoma, Biliary Stricture, Secondary Cholangitis, Jaundice.

INTRODUCTION

Biliary strictures most commonly involve the extrahepatic biliary tree [1], and typically present with clinical and biochemical signs of biliary obstruction [2]. Focal intrahepatic strictures (FIHS), affecting segmental intrahepatic ducts, are much rarer and are often asymptomatic [3].

There are numerous etiologies of biliary strictures, with malignancy whether primary or metastatic being the most common and clinically concerning cause. Such strictures are frequently detected incidentally during imaging performed for other indications [4].

The management of biliary strictures requires a thorough understanding of their natural history and an appropriate diagnostic evaluation. The two main priorities in patients presenting with biliary strictures are accurate diagnosis and biliary drainage specifically, confirmation or exclusion of malignancy and restoration of bile flow into the duodenum.

The objective of this case report is to describe the diagnostic approach and management of a benign left-sided biliary stricture.

CASE

A 64-year-old man with a history of left nephrectomy for renal tumor, cystoprostatectomy for bladder tumor, and appendectomy presented with right upper quadrant abdominal pain associated with jaundice and fever. Symptoms had been evolving for 25 days. Empirical antibiotic therapy had been initiated, and further investigations were performed.

Laboratory tests showed blood group A+, aspartate aminotransferase (AST) 112 IU/L (5–40), alanine aminotransferase (ALT) 200 IU/L (5–40), alkaline phosphatase 498 U/L (75–290), gamma-glutamyl transferase 156 IU/L (11–50), total bilirubin 14 mg/L (reference ≤10), direct bilirubin 9 mg/L (reference ≤5), albumin 31 g/L (35–52), prothrombin time 95%, white blood cell count 11.7 $\times 10^3/\mu L$ (4,000–10,000), hemoglobin 11 g/dL [11–16], and C-reactive protein 155 mg/L (0–5). Serologies for hepatitis B surface antigen,

HIV 1/2 antibodies, and hepatitis C antibodies were negative. Lipase and amylase levels were normal.

Abdominal ultrasound revealed marked dilatation of the left intrahepatic bile ducts without visible obstruction. Magnetic resonance imaging (MRI) demonstrated a short stenosis of the left hepatic duct without associated signal abnormality, with segmental dilatation of the left intrahepatic ducts containing multiple lithiasis. The right intrahepatic bile ducts were thin and non-dilated, without intraluminal lesions. The common bile duct was normal in caliber and without pathological findings. The gallbladder was acalculous with a thin wall.

Endoscopic retrograde cholangiopancreatography (ERCP)-guided biopsy was not feasible. After preoperative preparation, and with the patient classified as ASA II, a left hepatectomy was performed via laparotomy. The postoperative course was uneventful, with normalization of biological parameters. Histopathological examination confirmed the benign nature of the stricture, consistent with secondary lithiasic cholangitis.

Discussion

Primary sclerosing cholangitis (PSC) and secondary sclerosing cholangitis are characterized by inflammatory and fibrosing involvement of the intrahepatic and/or extrahepatic bile ducts. When a causative biliary injury is identified, the condition is referred to as secondary sclerosing cholangitis; in the absence of an identifiable cause, it is defined as primary sclerosing cholangitis. PSC is frequently associated with extrahepatic diseases, particularly inflammatory bowel disease (IBD), and more rarely with idiopathic, immune-mediated, or infiltrative disorders such as sarcoidosis [5].

Focal intrahepatic biliary strictures (FIHS) are often asymptomatic and are frequently discovered incidentally on imaging or at autopsy [7–9], making their true incidence difficult to determine. Approximately 10% of individuals without a history of PSC present PSC-like strictures at autopsy [9].

Clinically, patients with FIHS can be divided into two groups. About 30% are asymptomatic, with incidental discovery of the lesion. The remaining 60–70% present with nonspecific symptoms, mainly abdominal pain, transient fever, and jaundice [10]. This was the case in our patient, who presented with abdominal pain associated with jaundice and fever.

Previous reports have suggested that recurrent cholangitis is a predominant presentation. Conversely, other studies indicate that biliary obstruction involving up to 50% of the hepatic parenchyma may be well tolerated in the absence of infection and provided that the remaining liver is functional and non-obstructed. This was not the case in our patient, who presented with impaired liver function tests.

Benign biliary strictures may result from multiple pathological processes, including direct injury during biliary or hepatic surgery, hepatic arterial ischemia (e.g., after transarterial chemoembolization [0.3%] or radiofrequency ablation [17%]), chronic cholangitis (bacterial, eosinophilic, IgG4-related, or parasitic), and hepatolithiasis [11]. The main malignant cause of FIHS is cholangiocarcinoma, accounting for 50–53% of cases. Post-lithiasic inflammatory cholangitis complicated by infection was considered the most likely diagnosis in our patient. Malignant strictures are typically associated with significantly elevated CA 19-9 levels, whereas in our case, CA 19-9 was negative.

Ultrasound can detect intrahepatic bile duct dilatation (>2 mm) proximal to a stricture or vascular involvement [12]. However, in the absence of a mass, its ability to characterize strictures is limited [13, 14]. It may nevertheless detect associated findings such as hepatolithiasis [15], or cirrhosis. In our case, ultrasound did not reveal any mass.

Magnetic resonance imaging/magnetic resonance cholangiopancreatography (MRI/MRCP) detects biliary strictures with a sensitivity of 93.5% and a specificity of 94.4% [16]. It also allows assessment of vascular involvement. However, no study has definitively established its ability to distinguish benign from malignant intrahepatic strictures. In our case, MRI findings favored a benign stricture due to the absence of signal abnormality and the presence of a short-segment lesion.

Cholangioscopy provides direct visualization of intrahepatic duct lesions, allowing differentiation between benign and malignant conditions, early detection of multifocal disease, and targeted biopsies [17–22]. In our case, this procedure was not feasible, likely due to limited expertise and experience.

Endoscopic ultrasound and intraductal ultrasound may be useful when ERCP fails; however, these techniques were not available due to technical limitations. Consequently, cholangiocarcinoma could not be formally excluded preoperatively.

Conclusion

In the presence of a focal intrahepatic biliary stricture, malignancy should be considered first. Even a simple suspicion may lead to surgical resection, given the high prevalence of biliary cancers in this context and the difficulty of definitively excluding malignancy preoperatively.

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