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## Secondary sclerosing cholangitis from percutaneous transhepatic biliary drainage in a patient with gallbladder cancer after surgery: A case report



Zhongbao Tan, Qingqing Wang, Xuequn Mao, Rong Zou, Jian Zhang, Zhenhai Di, Huanjing Wang\*

Department of Interventional Radiology, Affiliated Hospital of Jiangsu University, Jiangsu, 212001, China

### ABSTRACT

Secondary sclerosing cholangitis (SSC) is a chronic cholestatic liver disease characterized by fibrosis and stricture of the bile ducts. SSC in association with multiple factors such as spontaneous choledochoduodenal fistula and metastatic gallbladder cancer has rarely been reported. However, to the best of our knowledge, reports of SSC after percutaneous transhepatic biliary drainage (PTBD), especially in cases with diffuse calcification of the bile duct walls, have not been reported. We report a case of SSC from PTBD in a patient with gallbladder cancer after surgery. The patient underwent external percutaneous biliary drainage for malignant bile duct obstruction after cholecystectomy. Repeated exchanges were performed at the first and the sixth month after PTBD using an internal and external drainage catheter. Two months after the third catheter exchange, findings of laboratory and imaging examinations were suggestive of SSC. The liver function tests of the patient were suggestive of cholestasis. Multidetector computed tomography showed diffuse calcification of the bile duct walls. Cholangiography showed intrahepatic biliary stenosis or dilatation.

### Introduction

Percutaneous transhepatic biliary drainage (PTBD) has been used extensively for relieving benign and malignant obstructive jaundice. Secondary sclerosing cholangitis (SSC) is a chronic cholestatic liver disease characterized by inflammation, fibrosis, and stricture of the bile duct.<sup>1</sup> SSC after PTBD has not been reported. We present a case of SSC after PTBD in a patient with diffuse calcification of the bile duct walls observed on the computed tomography (CT) image.

### Case presentation

A 33-year-old man received cholecystectomy for a space-occupying lesion of gall bladder. The pathological diagnosis was a moderately differentiated adenocarcinoma of gallbladder with lymph node metastasis (hepatic artery and omental tissue station). The patient received systemic chemotherapy at 1 month after the surgery. The patient had to stop chemotherapy and underwent external percutaneous biliary drainage for malignant bile duct obstruction at 2 months after the surgery. Repeated exchanges were performed at the first and the sixth month after PTBD using an 8.5F internal and external drainage catheter. The drainage catheter was washed with metronidazole injection in case of poor drainage or when the color of bile changed from yellow to green. Two months after the third catheter exchange, the color of bile changed

from yellow to green again.

The patient visited our hospital for treatment. The results of his routine blood tests, C-reactive protein (CRP), and liver function tests were as follows: total bilirubin (TB) 157.89 mg/L, direct bilirubin (DB) 134.1 μmol/L, alkaline phosphatase (ALP) 448.0 U/L, gamma glutamyl transpeptidase (GGT) 409.3 U/L, white blood cell (WBC) count  $9.6 \times 10^9/L$ , neutrophil ratio (NEU%) 78.5%, and CRP 45 mg/L. Subsequently, he went to a hospital in his town and received empiric anti-infective therapy with a second-generation cephalosporin for a week. However, very mild response was observed. Two weeks later, the patient presented with a persistent low-grade fever. Multidetector CT showed diffuse calcification of the bile duct walls (Fig. 1). The results of his routine blood tests, CRP, and liver function tests were as follows: TB 433.55 mg/L, DB 371.4 μmol/L, ALP 694.8 U/L, GGT 293.1 U/L, WBC  $15.1 \times 10^9/L$ , NEU 84.6%, and CRP 121.3 mg/L. Multidrug resistant bacteria were indicated by bacterial susceptibility. Cholangiography showed intra-hepatic bile duct stenosis and dilatation. The patient was admitted to our institute with a diagnosis of SSC. The drainage catheter was exchanged with a new 10.2F internal and external drainage catheter (Fig. 2).

### Discussion

Sclerosing cholangitis is a chronic disease characterized by stricture of the bile ducts and high levels of ALP and GGT. It is divided into

\* Corresponding author. Affiliated Hospital of Jiangsu University, No. 438, Jiefang Road, Zhenjiang, 212001, Jiangsu Province, China.

E-mail address: [doctorwhj@163.com](mailto:doctorwhj@163.com) (H. Wang).

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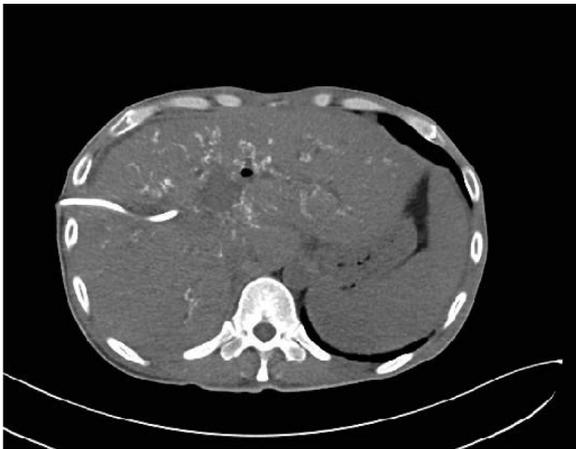


Fig. 1. Multidetector CT showed diffuse calcification of the bile duct walls.



Fig. 2. Cholangiography showed intra-hepatic bile duct stenosis and dilatation.

primary sclerosing cholangitis (PSC) and SSC. SSC is similar to PSC, but has innumerable causes such as infectious, immune-mediated, toxic, obstruction, choledochoduodenal fistulae, gallbladder carcinomas, and ischemic injury.<sup>1,2</sup> Two cases of spontaneous choledochoduodenal fistulae with repeated retrograde infection of bile duct were reported in the literature. These cases eventually developed into SCC.<sup>3,4</sup> Infection of biliary tract is one of the most common complications after PTBD. It is also an important cause of death, especially in patients with malignant tumor.<sup>5,6</sup>

Internal-external PTBD was conformed to physiological drainage. However, just like the spontaneous choledochoduodenal fistula, retrograde infections of the biliary tract are more likely to occur due to intestinal juice reflux through the internal and external drainage catheter, particularly in patients with malignant tumor.<sup>4,5</sup> In our patient, infection of the biliary tract after PTBD eventually lead to fibrosis, stricture, and calcification of the intra-hepatic bile duct, with subsequent development of SSC. SSC has also been reported to develop from a metastatic gallbladder carcinoma.<sup>7</sup> In the present case, PTBD was used for relieving symptoms of jaundice caused by lymphatic metastasis of gallbladder carcinoma. PTBD did not prevent the occurrence of SSC. Instead, recurrent infection of the biliary tract after PTBD facilitated the development of SSC.

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