An Unusual Case of COVID-19: What Is This 3D Printed Model of the Biliary Tree?

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Question: A 78-year-old woman presented with severe acute respiratory syndrome (SARS) owing to coronavirus disease 2019 (COVID-19). During the hospitalization, she developed abdominal pain, fever, and jaundice with leukocytosis (white blood count of 16×10^9/L), total bilirubin level of 21.1 mg/dL (reference range, 0.4–1.4 mg/dL), alkaline phosphatase of 870 U/L (reference range,30–135 U/L), and serum alanine aminotransferase of 77 U/L (reference range, 10–55 U/L).

An abdominal ultrasound examination showed no dilation in the intrahepatic bile duct. Magnetic resonance cholangiopancreatography revealed a dilated distal common bile duct with filling defects in the proximal common bile duct and tapering of the intrahepatic bile ducts concerning for cholangitis. Endoscopic retrograde cholangiopancreatography (ERCP) was subsequently performed, During the biliary sweep with a retrieval basket, an uncommon structure—similar to a 3-dimensional-printed model of the biliary tract (Figure *A*)—was removed. Final cholangiography demonstrated progressive destruction and obliteration of the intrahepatic bile ducts (pruned tree). A plastic biliary stent was then placed.

The procedure was uncomplicated with improvement in clinical symptoms and laboratory derangements. Owing to the active COVID-19 infection, the patient was eventually discharged after 120 days of hospitalization. Despite the diagnosis of secondary sclerosing cholangitis in critically ill patients (SSC-CIP), the patient was doing well at one-month follow-up with plan for repeat ERCP in 2 months.

What is the diagnosis?

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Conflicts of interest
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Answer to: Image 5: Biliary Cast Syndrome

Biliary cast syndrome is characterized by the development of casts in the biliary tree. Although uncommon, it is usually associated with liver transplantation.[1] However, critical illness owing to COVID-19 may result in biliary complications such as bile duct casts and progressive destruction of the intrahepatic bile ducts.

The etiology of cast development is not fully understood but is likely to be multifactorial, including sludge as a prerequisite and related factors such as bile duct damage and ischemia, biliary infection, hemolysis, fasting, parenteral nutrition, abdominal surgery, hypovolemic shock, and polytrauma.[1,2] Biliary cast syndrome is a hallmark of SSC-CIP. This is characterized by persistent cholestasis, biliary cast formation, and a stricturing fibrosing cholangiopathy.[2]

Different from our experience, endoscopic management has been shown to be effective in a minority of cases and surgical removal of casts is usually required.[1] However, SSC-CIP carries a high risk for hepatobiliary complications and, if progression toward cirrhosis occurs, the definitive treatment remains liver transplantation.[2]

Some cases of SSC-CIP among patients with SARS, especially owing to H1N1 influenza infection, have been reported; however, few reports have demonstrated a relationship to COVID-19.[2,3] The combined rarity of SSC-CIP associated with SARS-CoV2 infection, successful management by endoscopy, as well as the removal of the entire biliary casts as 1 piece during ERCP, as described here, adds a valuable contribution to the literature, especially given the COVID-19 pandemic.

Keywords: Endoscopy; Gastrointestinal; Bleeding; COVID-19; ECMO.

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