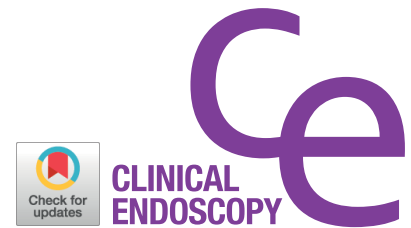


CASE REPORT

Clin Endosc 2021;54:285-288
https://doi.org/10.5946/ce.2020.167
Print ISSN 2234-2400 • On-line ISSN 2234-2443



Open Access

A Case of Congenital Common Bile Duct Web Treated with Balloon Dilatation under Endoscopic Retrograde Cholangiopancreatography in a Young Child

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Web in common bile duct (CBD web) is very rare. It is usually asymptomatic and detected incidentally during surgery for other causes in adults. It can be congenital or acquired, however congenital CBD web is extremely rare. Currently, despite its invasiveness and complications, endoscopic retrograde cholangiopancreatography (ERCP) is considered as a useful diagnostic and therapeutic modality in children with hepatobiliary pancreatic diseases as in adults. Herein we report a case of congenital CBD web presenting with acute pancreatitis and choledocholithiasis in a 4-year-old girl which was diagnosed and treated using balloon dilatation under ERCP. After balloon dilatation of the web, a common pancreatobiliary channel was observed. To the best of our knowledge, a case of congenital CBD web with pancreatobiliary junctional abnormality treated using ERCP in a child has not been reported to date. **Clin Endosc 2021;54:285-288**

Key Words: Common bile duct; Congenital; Endoscopic retrograde cholangiopancreatography; Web

INTRODUCTION

Extrahepatic biliary obstruction is often caused by a common bile duct (CBD) stone, a benign or malignant biliary stricture in adults¹; however, in children more diverse factors such as primary biliary cholangitis, post-liver transplantation, extrahepatic biliary atresia, and choledochal cyst are involved.^{2,3} CBD stone may cause extrahepatic biliary obstruction in children, but it is uncommon. In addition, congenital CBD web is an extremely rare cause of extrahepatic biliary

obstruction.⁴

Endoscopic retrograde cholangiopancreatography (ERCP) is widely used as a diagnostic and therapeutic modality for pancreaticobiliary disease (PBD) in adults. ERCP was less commonly performed in children because of the lower incidence of PBD in children than in adults; furthermore, the invasiveness nature of the procedure and the associated complications rendered it less suitable for children.³ However, the effectiveness of ERCP in the management of pediatric PBD has been reported recently.⁵⁻⁹

A small number of congenital biliary web cases in infants and children has been reported worldwide.⁴ However, to the best of our knowledge, a case of congenital web of the CBD in a child treated with balloon dilatation under ERCP has not yet been reported. Herein, we report a case of congenital CBD web diagnosed and treated with ERCP in a previously healthy 4-year-old girl.

This case report was reviewed and approved by the International Review Board of Gyeongsang National University Hospital (GNUH 2020-03-005).

Received: June 16, 2020 Revised: June 30, 2020

Accepted: July 1, 2020

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CASE REPORT

A 4-year-old girl visited our hospital because of colicky abdominal pain. The patient complained of abdominal pain 1 day ago. The pain was throbbing and localized in the peri-umbilical area. She refused to eat her meal due to aggravation of the abdominal pain and was seldom able to move. Fever, vomiting, or diarrhea was absent. Furthermore, no history of previous medical problem, surgery, or abdominal trauma was present. The mother had several silent stones in the gallbladder (GB), which were detected during routine health checkup.

The patient's weight and height were 18 kg (50–75th percentile) and 103.1 cm (50–75th percentile), respectively. Vital signs were relatively stable, with a blood pressure of 100/70 mm Hg, heart rate of 104 beats/min, respiratory rate of 28 breaths/min, and body temperature of 36.5°C. Physical examination revealed decreased bowel sound and abdominal tenderness on the periumbilical area.

The hemoglobin, white blood cell, and platelet counts were 13.1 g/dL, 8,210/mm³, and 310 × 10⁶/mm³, respectively.

Hemolysis was not noted. Blood chemistries were as follows: cholesterol 157 mg/dL (reference range, 120–200 mg/dL), total protein 7.9 mg/dL (6.1–7.9 mg/dL), albumin 4.9 mg/dL (3.5–5.6 mg/dL), alkaline phosphatase 294 U/L (134–386 U/L), aspartate aminotransferase (AST) 271 U/L (15–40 U/L), alanine aminotransferase level (ALT) 115 U/L (5–45 U/L), gamma glutamyl transpeptidase (GGT) 202 U/L (5–32 U/L), total bilirubin 0.63 mg/dL (<2.0 mg/dL), amylase 320 U/L (16–91 U/L), and lipase of 567 mg/dL (4–29 mg/dL).

Abdominal X-ray showed focal ileus in the central part of the abdomen. Abdominal ultrasonography (USG) revealed a swollen pancreas, dilated intrahepatic bile duct, and a CBD 0.94 cm in diameter and a 4.7-mm-sized echogenic focus in the distal CBD (Fig. 1A, B). The echogenic focus was suggested to be a CBD stone without posterior shadow. A septum in the CBD was noted on the coronal view of pancreaticobiliary computed tomography (CT; Fig. 1C). Acute pancreatitis, with biliary obstruction due to the stone and web-like structure in the CBD, was diagnosed. Non per oral diet, intravenous hydration, pain killers, and antibiotics (cefotaxime and metroni-

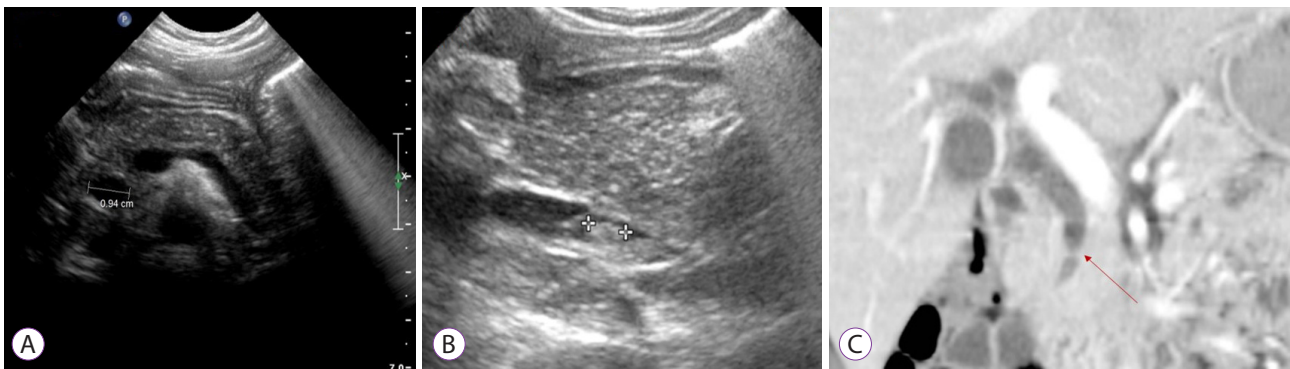


Fig. 1. Abdominal radiologic studies show the dilated intrahepatic and common bile duct (CBD) with 0.94 cm in diameter (A), and a 4.7-mm-sized echogenic material in the distal CBD (B) on abdominal ultrasonography. Pancreaticobiliary computed tomography reveals a web-like structure (arrow) in the CBD (C).

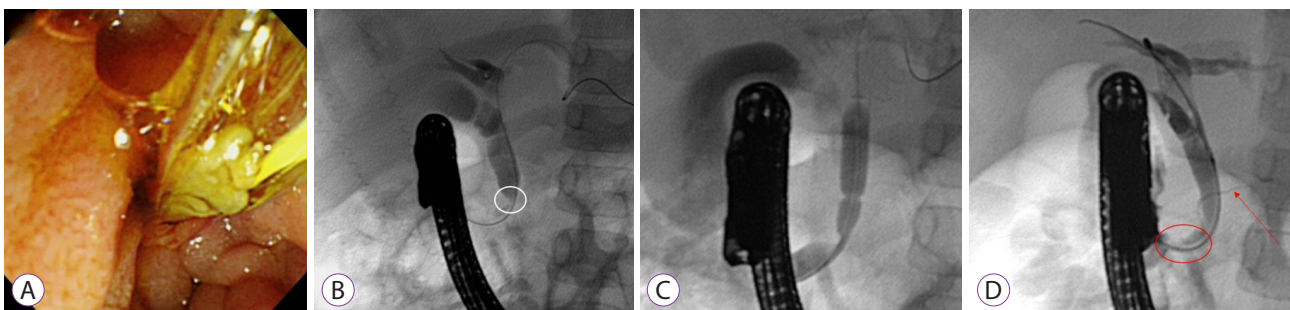


Fig. 2. Squashed muddy stone from the common bile duct (CBD) is extracted by endoscopic balloon dilation (A). Endoscopic retrograde cholangiography shows dilated proximal bile ducts and a narrowing and a shelf-like linear contrast shadow (white circle) in the distal CBD (B). Balloon dilation of the bile duct is performed to dilate the orifice and stricture of the CBD (C). Cholangiography after CBD dilatation shows dilated stricture and a common channel (red circle) with the pancreatic duct (red arrow, D). Abnormal pancreaticobiliary junction is noted.

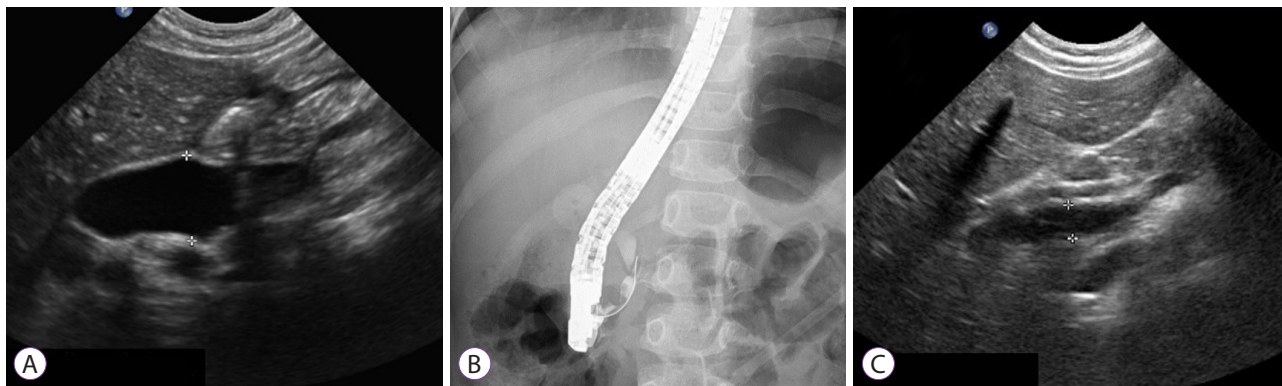


Fig. 3. Marked dilated intrahepatic and extrahepatic duct (1.63 cm in diameter) is shown on abdominal ultrasonography, when the patient was re-admitted (A). Endoscopic retrograde cholangiography shows a tight stricture and filling defects in the common bile duct (B). After recanalization, the dilated extrahepatic duct is improved to 0.67 cm in diameter (C).

dazole) were started. We decided to perform ERCP to evaluate and manage the stones and septum in the CBD. Endoscopic procedure was performed on the 6th day of admission under general anesthesia when the serum amylase and lipase levels decreased to 75 U/L and 32 mg/dL, respectively. A squashed muddy stone was extracted from the CBD (Fig. 2A). A thin narrowing and a shelf-like linear contrast shadow in the distal CBD as well as proximal bile duct dilatation were observed on ERCP (Fig. 2B). The CBD patency was re-established by balloon dilation (Fig. 2C), and an abnormal pancreaticobiliary junction of the common channel was suspected (Fig. 2D). Post-ERCP pancreatitis occurred, and the serum amylase and lipase levels increased to 1,749 U/L and >700 mg/dL, respectively. Conservative treatment was started again. After regression of the post-ERCP pancreatitis, the patient was discharged in good general condition on the 15th day of hospitalization. After 11 months, the patient visited our hospital again due to pancreatitis and biliary tract dilation caused by distal CBD stricture, probably due to previously incomplete recanalization (Fig. 3A, B). ERCP with balloon dilation was performed, and the child was discharged on the 5th day of re-hospitalization with improvement of pancreatitis and distal CBD stricture (Fig. 3C). She has been doing well for 4 years since then.

DISCUSSION

CBD web is caused by congenital malformation and acquired inflammation due to recurrent stones or injury, but it is uncommon. To date, a number of CBD web cases in adults has been reported worldwide; however, a pediatric case of congenital CBD web is rare.^{1,4,10-13} Acquired CBD web is caused by injury during biliary procedure or surgery, chronic inflammation due to recurrent stones, and malignancy.^{2,13} Congenital

CBD web, on the other hand, is caused by incomplete biliary tract recanalization during embryogenesis,¹² and it is often accompanied by a choledochal cyst, hepatic fibrosis, and anomalous junction of the pancreaticobiliary ductal system.^{4,12} In our patient, CBD stricture may be congenital rather than acquired because she is young and previously healthy, and surgery or trauma is absent. In addition, the external CBD shape is preserved, and the concomitant muddy and soft CBD stone may not cause biliary tract irritation.

CBD web can present at any age depending on the degree of ductal patency for bile flow,^{4,12} although it is usually asymptomatic. CBD web may lead to obstructive jaundice associated with cholangitis or cholelithiasis.⁴ In the patient, acute pancreatitis was presented rather than obstructive jaundice, which might have been caused by the associated pancreaticobiliary maljunction. Biliary flow might be relatively preserved because of the incomplete CBD obstruction by the web and muddy stone. Although the CBD stone and web did not cause obstructive jaundice in the patient, they caused hepatocellular and canalicular injuries judging from the elevated AST, ALT, and GGT on the chemical analyses.

The CBD web diagnosis is challenging because it is often asymptomatic and detected by chance during ERCP or surgery.¹⁰ Sometimes, excessive contrast media injection into the biliary tract during ERCP can result in an image similar to that of the CBD web; however, it is usually resolved by timely spontaneous excretion.¹¹ Abdominal USG or CT can easily detect the dilated bile ducts; however, it is challenging to detect any membranous structure in the biliary ducts.¹² In the patient, the stones and the dilated biliary tract were detected on abdominal USG and CT, and the coronal view of pancreaticobiliary CT suggested a membranous structure in the CBD (Fig. 1C).

Surgical resection is the treatment of choice in CBD web with cholestasis or cholangitis.¹¹ However, successful endo-

scopic balloon dilation and stent insertion in adults has been reported in Korea.^{10,14} Endoscopic treatment can be considered in a case of incomplete or mild form of obstruction,¹² and ERCP in the pediatric population is considered as a safe and efficient diagnostic and therapeutic modality in PBD.^{6,9,15}

In our case, the endoscopic approach was considered for the treatment of CBD stone and web after acute pancreatitis regression. Endoscopic CBD stone removal and balloon dilation of the CBD web were successfully performed. After re-establishing the CBD patency, a common biliary channel with the pancreatic duct was noted on cholangiography (Fig. 2D).

ERCP-related pancreatitis is not uncommon.³ Unfortunately, pancreatitis occurred after ERCP in the patient, which was caused by pancreaticobiliary maljunction, previous pancreatitis, and ERCP itself.

CBD stone is rare in children, although it has become more common nowadays. Most of the stones in children are black pigment or cholesterol stones and usually originate from the gall bladder (GB).¹⁶ In the patient, an isolated CBD stone without gallstone was detected, and it was soft, greasy, and bright yellow. Based on the clinical information, the CBD stone was caused by the CBD web and the pancreaticobiliary junctional abnormality. Hemolysis or the use of drugs, such as ceftriaxone, was not noted. Since the incidence of GB stone in adults is relatively high, although wide variations among racial and ethnic groups exist,¹⁶ genetic background for the CBD stone in this patient is less likely, despite the mother having an asymptomatic GB stone. The CBD web and stone have been endoscopically removed, but the patient still has a risk for CBD stone formation and pancreatitis occurrence owing to her pancreaticobiliary junctional abnormality. Therefore, regular and serial check-ups are warranted.

Previously, a congenital CBD web associated with hepatobiliary pancreatic ductal anomalies in an 8-year-old boy was reported in India. However, to the best of our knowledge, a congenital CBD web case in early childhood successfully treated by balloon dilation under ERCP has not been reported.¹⁷ Although it is rare, congenital CBD web could be one of the differential diagnoses of choledocholithiasis and acute pancreatitis in children.

Conflicts of Interest

The authors have no potential conflicts of interest.

Funding

None.

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