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Single-center experience of choledochoscopy in pediatric patients

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Choledochoscopy has transformed the field of pancreatobiliary endoscopy by allowing direct visualization of the biliary tree, selective biopsy, and destruction of intraductal stones. The technique was developed 4 decades ago and initially was used only intraoperatively or through established percutaneous tracks.^{1,2} In the 1970s, Kawai et al³ adapted the technique to be used in conjunction with duodenoscopy. This advancement provided a single endoscopic solution to 2 important problems in the field: (1) indirect imaging of the biliary and pancreatic tree obtained by available techniques, such as ERCP, and (2) inability to sample ductal tissue, unless through invasive surgical procedures or less-sensitive intraductal brushings.

Despite these advantages, choledochoscopy was limited by several technical considerations that continue to be improved in an iterative process. First, the original technique depended on 2 operators, 1 to maneuver the “mother” duodenoscope and the second to operate the miniscope.^{1,4} Now, single-operator choledochoscope systems exist, with both scopes manipulated by 1 endoscopist.^{5,6} Additional shortcomings of the early-generation miniscopes included expense, fragility, and poor optics. Furthermore, they were difficult to maneuver and had insufficient tip deflection to enable visualization of all parts of the lumen. Newer miniscopes have improved on many of these problems, including improved imaging technology, 4-way deflected steering, and a separate channel for water irrigation. The improvements have led to greater use of choledochoscopy in the evaluation of pancreaticobiliary disease.

In pediatric populations, choledochoscopy still remains a relatively unused technology compared with ERCP. Siddique et al² reported using a percutaneous approach in a 3-year-old child, but otherwise, to our knowledge, no other reports or case series exist in the

literature. Since 2008 we have adapted choledochoscopy as part of the repertoire of biliary imaging at our center. In this report, we describe the largest pediatric experience with choledochoscopy, outlining the indications and outcomes.

METHODS

We retrospectively reviewed the charts of all patients undergoing choledochoscopy through oral or percutaneous transhepatic catheter (PTC) routes. Indications included abnormal imaging (ERCP or magnetic retrograde cholangiopancreatography [MRCP]), abnormal liver panel, obstructive jaundice, or cancer surveillance. Procedures were performed with the patients under modified or general anesthesia by an experienced biliary endoscopist (I.R. or D.F.). All patients undergoing peroral procedures had a sphincterotomy (either before or at the time of endoscopy) and received prophylactic antibiotics. This study was approved by the Institutional Review Board at Baylor College of Medicine.

Two choledochoscopes were used in these cases. The majority used the Spyglass Spyscope (Boston Scientific, Natick, MA), which has a 10F diameter, 4-way tip deflection, 1.2-mm working channel, and a separate channel for water irrigation. The Spyscope apparatus was advanced through the Pentax 3490T (Pentax, Montvale, NJ) duodenoscope and then into the bile duct, with the guidewire removed to facilitate tip deflection (Fig. 1). In addition to the Spyscope, the Olympus SIF-180 (Olympus, Center Valley, PA) enteroscope was also used in 1 patient with exceptionally dilated bile ducts. The Olympus SIF-180, which has a 9.2-mm distal diameter and a 2.8-mm working channel, was advanced directly through a Rouxen-Y limb into the hepatobiliary system (Fig. 2, Table 1).

RESULTS

Eleven patients underwent intraductal biliary evaluation. The youngest patient was 2 years old, and the median age was 14 years. The patients presented with a variety of underlying diagnoses, including primary sclerosing cholangitis ($n = 3$), history of common bile duct stones ($n = 3$), and history of lymphoma ($n = 1$). Two patients had undergone orthotopic liver transplantation (OLT). The most common indication for choledochoscopy was suspected obstruction (in 10 of 11 patients), determined by abnormal bilirubin levels and imaging. The 1 patient with normal bilirubin levels had significantly elevated aminotransferases and markedly dilated intrahepatic bile ducts after hepatoporojejunostomy for a choledochal cyst.

Visualization of biliary mucosa was possible in all cases. In 9 cases, the choledochoscope was inserted perorally. In the other 2 cases, a preexisting percutaneous transhepatic catheter was used as a portal through which to pass the endoscope. Three patients had mucosa that appeared normal without any signs of disease or obstruction. Stone disease was confirmed in 2 patients and biliary stricture in 2 patients. Biopsy specimens were obtained in 2 cases, with 1 demonstrating marked eosinophilia in ductular tissue despite an endoscopically normal appearance on cholangioscopy.

Choledochoscopy altered management in the majority of the cases. In the relevant cases, a normal appearance or negative biopsy result was able to rule out cholangiocarcinoma,

lymphoma, or other mucosal cause of bile duct obstruction. Furthermore, in 2 cases in which obstruction could not be relieved or in 1 case (case 3) where intrahepatic disease was evident without a cause of extrahepatic disease, patients were listed for liver transplantation. One case confirmed a complicated bile duct stenosis (case 7) seen on magnetic resonance imaging, and with direct visualization, cannulation was possible. A biliary stent was placed to bypass the obstruction until a more definitive hepatojejunostomy was performed. Finally, in the case with eosinophils found on biopsy, steroids were given, and the patient's symptoms and laboratory test results improved.

Few patients had side effects directly attributable to choledochoscopy. There were no postoperative episodes of pancreatitis, although 1 patient had abdominal pain with normal lipase levels, which resolved over 12 hours. One patient with suspected malignant obstruction experienced bacteremia, requiring intensive care, likely secondary to the underlying condition (chemotherapy-induced neutropenia) and periprocedural seeding.

DISCUSSION

Evaluation of the biliary tree in pediatric liver disease has largely been accomplished by indirect methods. Damage to the biliary epithelium is suggested by abnormal laboratory values, such as alkaline phosphatase and gamma-glutamyl transpeptidase, but the values are neither sensitive nor specific. Biliary structures can be visualized through noninvasive imaging techniques, such as abdominal US or MRCP, which can detect common bile duct dilation, stones, or tumors. ERCP and intraoperative cholangiogram provide an outline of the biliary tree with therapeutic options, but the combined use of choledochoscopy can provide direct visualization or sampling of the tissue.

In this series, we demonstrate that choledochoscopy is a powerful tool for diagnosing and treating a variety of pediatric biliary diseases. The choledochoscope can be inserted perorally or transcutaneously through a PTC, and it can provide visualization and biopsy of biliary tissues. The procedure can be used for several purposes, including stone detection, evaluation for obstructing tumor, and examination of the anastomosis site after surgical duct-to-duct ligation. The procedure has the added advantage of carrying risks similar to those of ERCP or other endoscopic procedures, with only 1 case complicated by bacteremia in an already immunocompromised host.

Choledochoscopy should be considered as a tool to integrate into practice, inasmuch as it significantly altered the care plan for the patients in this study. Choledochoscopy changed management in 7 of 11 of our cases, including listing for liver transplantation ($n = 2$), keeping/removing the PTC ($n = 2$), referring for hepatojejunostomy ($n = 1$), deferring further surgery ($n = 1$), and initiating new medical management ($n = 1$). Furthermore, in 4 of 11 cases an intervention was performed—assisted wire cannulation ($n = 2$) and stone removal with direct visualization ($n = 2$). Choledochoscopy was used for management before and after liver transplantation in 6 of 11 patients. Further studies, with larger numbers of patients, will better define the uses of choledochoscopy, including emerging uses of lithotripsy in pediatric choledocholithiasis.

The future also promises further technologic improvements to make the choledochoscopes easier to use. The optics are still improving to acquire images similar to those obtained by our current high-definition endoscopes. Additionally, there are limitations for use specifically in children because of the choledochoscope diameter and channel size, which can limit the number of simultaneous therapies possible. These shortcomings will likely be addressed in the near future as pediatric gastroenterologists gain more experience with—and discover new uses for—choledochoscopy.

Abbreviations

MRCP	magnetic retrograde cholangiopancreatography
OLT	orthotopic liver transplantation
PTC	percutaneous transhepatic catheter

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Figure 1.
A, Cholangiogram demonstrating intraductal cannulation of choledochoscope in a 3-year-old child. B, Direct visualization of biliary mucosa (patient 8).

Figure 2.
A, Endoscopic view of biliary mucosa and biopsy forceps in a 4-year-old child. **B,** Histologic appearance demonstrating marked eosinophilia in biliary mucosa (patient 6) (H&E, orig. mag. $\times 100$).

TABLE 1

Case information

Case	Age (y)	Diagnosis	Indication	Findings	Management change	Complications
1		PSC	<ul style="list-style-type: none"> Evaluate for cholangiocarcinoma vs other obstructive cause 	<ul style="list-style-type: none"> No obstruction seen Biopsy specimen negative for cholangiocarcinoma 	Removed PTC	None
2	14	PSC	<ul style="list-style-type: none"> Abnormally shaped common hepatic/cystic duct junction Evaluate for obstruction 	<ul style="list-style-type: none"> Unable to visualize mucosa at site of obstruction 	None	None
3	15	Hyper IgM syndrome (PSC)	<ul style="list-style-type: none"> Progressive jaundice Abnormal liver histology Persistent extrahepatic ductal dilation Evaluate for obstruction or infection 	<ul style="list-style-type: none"> Normal biliary mucosa No identifiable extrahepatic obstruction 	Listed for liver transplant	Abdominal pain, normal lipase
4	2	Post-OLT lymphoma	<ul style="list-style-type: none"> Evaluate for tumor obstruction vs postoperative duct stricture 	<ul style="list-style-type: none"> Areas of mucosa with mild inflammation No evidence of lymphoma 	Placed PTC	Bacteremia
5	21	PSC	<ul style="list-style-type: none"> Cannulate obstruction seen on ERCP 	<ul style="list-style-type: none"> Unable to cannulate obstruction 	Listed for liver transplant	None
6	4	Choledochal cyst	<ul style="list-style-type: none"> Persistent cholangitis 	<ul style="list-style-type: none"> Biopsy specimen positive for eosinophils 	Initiated IV steroids	None
7	18	Postcholecystectomy injury	<ul style="list-style-type: none"> Cannulate obstruction 	<ul style="list-style-type: none"> Placed stent 	Referred for hepaticojejunostomy	None
8	3	Dilated CBD	<ul style="list-style-type: none"> Evaluate for obstructive mass Possible lithotripsy 	<ul style="list-style-type: none"> No masses Stone debris seen 	Deferred surgery	None
9	11	Retained CBD stones after cholecystectomy	<ul style="list-style-type: none"> Evaluate for stones Possible lithotripsy 	<ul style="list-style-type: none"> Stone debris removed Nonobstructive stricture identified 	None	None

Case	Age (y)	Diagnosis	Indication	Findings	Management change	Complications
10	14	Post-OLT biliary stricture	<ul style="list-style-type: none">Evaluate obstruction	<ul style="list-style-type: none">Scarring of extrahepatic ductular mucosa	None	None
11	14	Retained CBD stones after cholecystectomy	<ul style="list-style-type: none">Evaluate for stonesPossible lithotripsy	<ul style="list-style-type: none">No stones visualizedNormal biliary mucosa	None	None

CBD, Common bile duct; *IgM*, gamma M immunoglobulin; *IV*, intravenous; *OLT*, orthotopic liver transplantation; *PCT*, percutaneous transhepatic catheter; *PSC*, primary sclerosing cholangitis.