Pediatr Surg Int (2007) 23:111–116
DOI 10.1007/s00383-006-1832-3

ORIGINAL ARTICLE

Diagnostic and therapeutic ERCP in the pediatric age group

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Received: 5 October 2006 / Accepted: 16 October 2006 / Published online: 6 December 2006

Abstract The role and value of endoscopic retrograde cholangiopancreatography (ERCP) in the pediatric age group is not well established, because pancreatic and biliary diseases are less common in children. This however is not the case in areas like the Eastern Province of Saudi Arabia where sickle cell disease (SCD) and other hemoglobinopathies are common, with increased frequency of cholelithiasis and choledocholithiasis. The purpose of this study was to evaluate the indications, findings, safety and therapies of ERCP in children. One hundred and twenty five children had diagnostic and/or therapeutic ERCP as part of their management at our hospital. Their medical records were reviewed for: age at diagnosis, sex, Hb electrophoresis, indication for ERCP, findings, therapy and complications. There were 77 males and 48 females. Their age at presentation ranged from 5–18 year (mean 13.25 year). The majority of them had sickle cell disease (77.6%). The indications for ERCP were: obstructive jaundice (67.2%), recurrent biliary colic with or without jaundice (10.4%), acute and chronic pancreatitis (7.2%), postoperative bile leak (2.4%), cholangitis with obstructive jaundice (2.4%), hepatitis of unknown etiology (3.2%), cirrhosis of unknown etiology (4%), thalassemia with jaundice (0.8%), hemobilia (0.8%), acute cholecystitis with jaundice (0.8%), and sickle cell disease with ulcerative colitis and obstructive jaundice (0.8%). In six children, ERCP was done following laparoscopic cholecystectomy. ERCP was carried out under sedation in 91 (72.8%) children and under general anesthesia in 34. It was successful in 121 (96.8%) children while cannulation of the Ampulla failed in four. ERCP was normal in 43 children, but eight of them showed evidence of recent stone passage and in six, there were gallstones. In the remaining children, ERCP revealed: normal CBD with stones (18 patients), dilated CBD with stones (17 patients), dilated CBD without stones (19 patients), dilated biliary tree with stones (10 patients), dilated biliary tree without stones (six patients), bile leak (two patients), dilated biliary tree with stones and choledocho-duodenal fistula (one patient), choledochal cyst (two patients), septate gallbladder (one patient), normal ERCP with multiple pancreatic cysts (one patient) and biliary stricture (one patient). The following procedures were carried out: 35 had endoscopic sphincterotomy and stone extraction, 20 had endoscopic sphincterotomy, four had CBD stenting, one underwent removal of a stent, two had insertion of a nasobiliary tube and one had biliary endoprosethesis. There was no mortality. One had bleeding from the site of sphincterotomy which stopped after adrenaline injection. Four patients (3.2%) developed transient mild pancreatitis which settled conservatively. ERCP in the pediatric age group is safe both as a diagnostic and therapeutic procedure. ERCP can provide valuable information which aid in the diagnosis of biliary and pancreatic diseases in children as well as therapy with the technical feasibility of endoscopic sphincterotomy. This is specially so in the era of laparoscopic cholecystectomy, where ERCP should be the treat-

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ment of choice in children with CBD stones who are going or have previously undergone laparoscopic cholecystectomy.

Keywords Endoscopic retrograde cholangiopancreatography · Children · Endoscopic sphincterotomy · Cholelithiasis · Choledocholithiasis · Sickle cell disease

Introduction

Endoscopic retrograde cholangiopancreatography (ERCP) was first described in 1970 by Demling and Classen [1]. Since then, it has become the procedure of choice for the diagnosis and treatment of biliary and pancreatic diseases in adults. In the pediatric age group, the value of ERCP is not well defined. This is attributed to a low prevalence of pancreaticobiliary diseases in children and the few earlier reports in the literature described ERCP as a diagnostic investigation in children [2–4]. The recent liberal use of ultrasound for the investigation of children with abdominal complaints resulted in an increase detection of biliary and pancreatic diseases. This is more so in areas like the Eastern Province of Saudi Arabia where hemoglobinopathies are common with increased frequency of cholelithiasis and choledocholithiasis [5]. As a result of this, ERCP is being used with increased frequency for the investigation as well as treatment of biliary and pancreatic diseases in children [6–8]. This is specially so in the era of laparoscopic cholecystectomy where ERCP and endoscopic sphincterotomy have become the treatment of choice for bile duct stones whether preoperatively or postoperatively [9, 10]. This report describes our experience with ERCP as a diagnostic and therapeutic procedure in 125 children with biliary and pancreatic disorders.

Patients and methods

From September 1993 to August 2005, ERCP was performed for 125 children less than 18 year old as part of their management at our hospital. Their medical records were retrospectively reviewed for: age at diagnosis, sex, indication for ERCP, preoperative investigations including Hb electrophoresis for those with sickle cell disease, ERCP findings and procedures, complications, and post-ERCP management. Children with SCD were properly hydrated with intravenous fluids starting the night before the procedure at a rate of 1½ their maintenance requirements and when necessary given blood transfusions to increase their Hb to 10–12 g/dl.

All ERCPs were performed in the radiology department using Olympus JF1 T20 side-viewing dudenoscope. This was done under general anesthesia with nasotracheal intubations for children less than 10 years old, and under sedation using meperdine (1 mg/kg) and diazepam (0.1–0.2 mg/kg) for those above 10 years of age. The ampulla's of Vater was cannulated with tapered catheters and the pancreatic and biliary ducts were visualized by fluoroscopy using Hexabrix (320 mg diluted to 50%). Appropriate radiographs were obtained and where indicated sphincterotomy was performed using 5F sphincterotome (Olympus) and common bile duct stones if found were extracted with a basket and balloon catheters.

Results

One hundred and twenty five children had ERCP as part of their management at our hospital. There were 77 males and 48 females (M:F ratio of 1.6:1). Their age at presentation ranged from 5–18 year (mean 13.25 year). The majority of them had SCD (97 (77.6%)) patients. Their mean Hb S level was 78% (range 65–90%) and their mean Hb F level was 22% (range 7–35%). The indications for ERCP are shown in Table 1. Obstructive jaundice was the commonest indication in 84 (66.4%) children, followed by recurrent biliary colic in 13 patients (11.2%). Acute and chronic pancreatitis was the indication in nine (7.2%) and three children had bile leak, post-traumatic in two and post- laparoscopic cholecystectomy in one. In six children, ERCP was done following laparoscopic cholecystectomy, in five because of obstructive jaundice and in one because of bile leak. Five children had liver cirrhosis of unknown etiology and four had hepatitis of unknown etiology. Three children had cholangitis and one each had hemobilia, acute cholecystitis with obstructive jaundice, thalassemia with obstructive jaundice, and SCD with ulcerative colitis and obstructive jaundice.

ERCP was done under sedation in 91 (72.8%) children and under general anesthesia in 34 (27.2%). The procedure was successful in 121 children (96.8%) and cannulation of the Ampulla failed in four. The findings during ERCP are shown in Table 2. ERCP was normal in 43 children, but eight of them showed evidence of recent stone passage (red, inflamed, edematous papilla) and six of them had gallstones. In the remaining patients, ERCP revealed normal CBD with CBD

Table 1 Indications for ERCP

Indication for ERCP	No. of patients	%
Obstructive jaundice	84	67.2
Recurrent biliary colic	13	10.4
Pancreatitis	9	7.2
Cirrhosis of unknown etiology	5	4
Hepatitis of unknown etiology	4	3.2
Bile leak	3	2.4
Cholangitis	3	2.4
Hemobilia	1	0.8
Acute cholecystitis with obstructive jaundice	1	0.8
Thalassemia major with obstructive jaundice	1	0.8
Sickle cell disease with ulcerative colitis and obstructive jaundice	1	0.8

Table 2 Findings during ERCP

Finding during ERCP	No. of children	%
Normal ERCP	43	34.4
Failed ERCP	4	3.2
Normal CBD with CBD stones	17	13.6
Dilated CBD with stones	17	13.6
Dilated CBD without stones	19	15.2
Dilated biliary ducts with stones	10	8
Dilated biliary ducts without stones	6	4.8
Bile leak (one of them had associated CBD stone)	3	2.4
Dilated biliary ducts with stones and choledocho-duodenal fistula	1	0.8
Choledochal cyst	2	1.6
Septate gallbladder	1	0.8
Multiple pancreatic cysts	1	0.8
Biliary stricture	1	0.8

stones in 18 (Fig. 1). In five of them there were gallstones, and in two there was associated biliary sludge in the CBD. Three of them had cholangitis and in one, there was pus in the CBD. One child had bile leak from the cystic duct stump following laparoscopic cholecystectomy and ERCP revealed a retained stone in CBD. The leak stopped after endoscopic sphincterotomy and stone extraction. Seventeen children had CBD dilatation and stones (Fig. 2). In three, gallstones were visualized and in three there was associated biliary sludge in the CBD. Dilated CBD without stones was seen in 19 children, in nine of them gallstones were visualized and in six, there was associated CBD biliary sludge. Ten children had dilated biliary ducts with stones and six had dilated biliary ducts but no stones (Fig. 3). Two children had liver injury with post-traumatic bile leak that settled following endoscopic sphincterotomy. One child had dilated biliary ducts

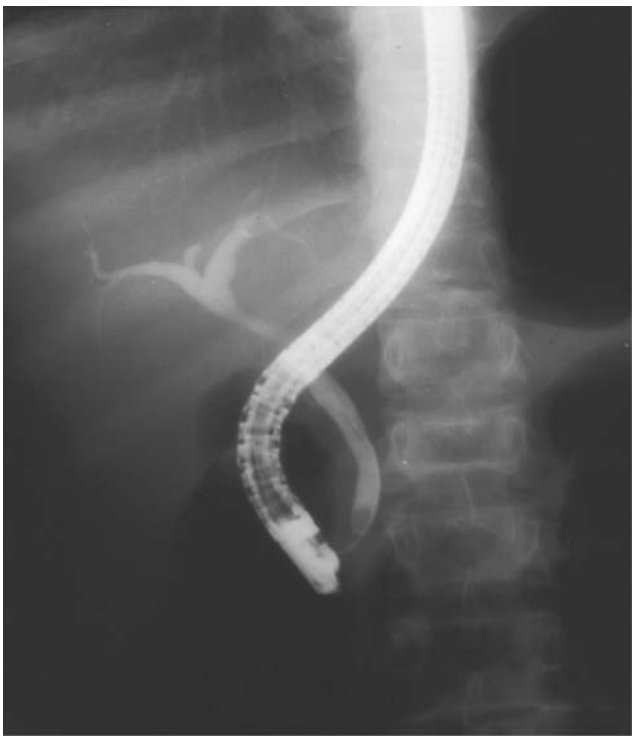

Fig. 1 ERCP showing normal CBD with a stone in the lower end

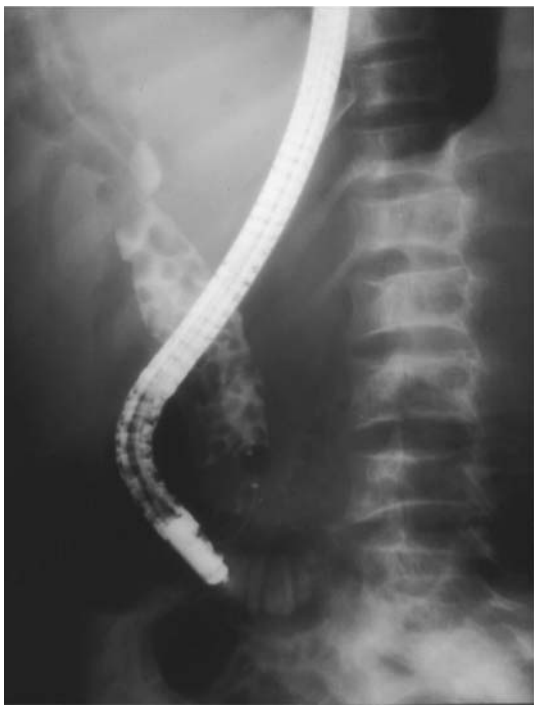

Fig. 2 ERCP showing dilated CBD with multiple stones

with stones and choledocho-duodenal fistula. Two children had features of choledochal cysts, one child had septate gallbladder and another child had multiple

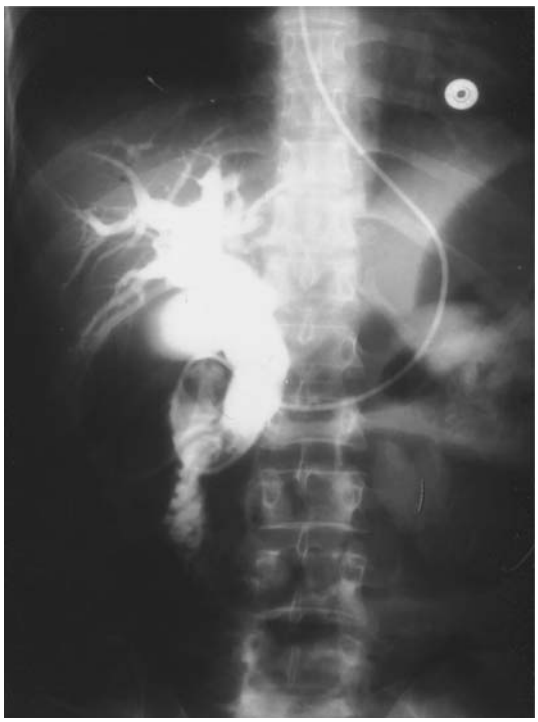

Fig. 3 ERCP showing dilated biliary ducts with no stones

pancreatic cysts. One child with SCD and ulcerative colitis had biliary stricture.

The different procedures performed during ERCP are shown in Table 3. Thirty-five had endoscopic sphincterotomy and stone extraction. In one, this was following laparoscopic cholecystectomy with a retained CBD stone. One of them had mechanical lithotripsy via ERCP and stone extraction because of the large size of CBD stone. Thirty-four of them subsequently had laparoscopic cholecystectomy. Twenty had endoscopic sphincterotomy only, four had endoscopic sphincterotomy and CBD stenting, and one underwent removal of a previously inserted stent. Two had endoscopic sphincterotomy and insertion of a nasobiliary tube and one had biliary endoprothesis.

Table 3 Therapeutic procedures performed during ERCP

Therapeutic procedure	No. of patients	%
Endoscopic sphincterotomy and stone extraction	35	28.9
Endoscopic sphincterotomy only	20	16.5
Endoscopic sphincterotomy and CBD stenting	4	3.3
Endoscopic sphincterotomy and insertion of nasobiliary tube	2	1.6
Endoscopic sphincterotomy and insertion of biliary endoprothesis	1	0.8
Removal of a eviously inserted stent	1	0.8

There was no mortality. One of our patients had bleeding at the time of sphincterotomy which stopped after adrenaline injection. Four children (3.2%) developed transient mild pancreatitis which settled conservatively.

Discussion

Endoscopic retrograde cholangiopancreatography is extensively used in adults. In the pediatric age group, ERCP is being used with increasing frequency as a diagnostic procedure for the evaluation of pancreatic and biliary diseases, and recently as a therapeutic procedure [2–4, 6–8]. The number of patients in published series remained however small. The main reason for this is that the number of children for whom the procedure is indicated remains small. Recently, there were few reported series describing ERCP in a relatively large number of children [11–13]. Our series is comparatively large, and one reason for this is the large number of children with SCD (77.6%). SCD is one of the commonest hemoglobinopathies in the Eastern Province of Saudi Arabia with a sickle cell trait frequency of about 25% in some areas [14]. Cholelithiasis and choledocholithiasis are common complications of SCD. The prevalence of cholelithiasis in patients with SCD is variable ranging from 17% to 55%, but the frequency increases with age [15–17]. In the Eastern Province of Saudi Arabia, an overall 19.7% frequency of cholelithiasis was reported in children with SCD. This frequency however, increased to 36% in those 15–18 years of age [5]. A 30% incidence of common bile duct stones was also reported in children with SCD undergoing cholecystectomy [18]. In the past and based on this high incidence of CBD stones, routine intraoperative cholangiogram was recommended during cholecystectomy as this may necessitate CBD exploration [18]. This however is not the case in the era of laparoscopic cholecystectomy. We found ERCP valuable both preoperatively and postoperatively, not only as a diagnostic but also as a therapeutic procedure. Six of our patients had ERCP post laparoscopic cholecystectomy, in five because of obstructive jaundice, while in the sixth it was because of bile leak from the cystic duct stump. Three of them had retained common bile duct stones, one had stenosis of the papilla, and the other two had dilated biliary ducts but no stones. In all, ERCP proved valuable. Thirty-six of our patients had ERCP with sphincterotomy and stone extraction and

34 of them subsequently underwent laparoscopic cholecystectomy. This sequential approach of endoscpic sphincterotomy and stone extraction followed by laparoscopic cholecystectomy is a safe and effective approach for the management of children with cholelithiasis and chledocholithiasis [10]. Since we started using ERCP, none of our patients required CBD exploration and all CBD stones whether diagnosed preoperatively or postoperatively were managed by endoscopic sphincterotomy and stone extraction. Of interest was the finding of dilated bile ducts without evidence of obstruction in 26 of our SCD patients. The cause of this dilation is not known. All had endoscopic sphincterotmy, and four of them had CBD stenting, one also had a nasobiliary tube and another had biliary endoprothesis. The value of endoscopic sphincterotomy in these patients is not known, but it may prove to be useful in the future as they are liable to develop recurrent bile duct stones or sludge. This is more likely in the presence of dilated bile ducts and since these stones are small to start with, they as well as biliary sludge are likely to pass spontaneously in the presence of sphincterotomy. Biliary sludge is a mixture of calcium bilirubinate and cholesterol crystals within viscous bile that contains a high concentration of mucus and proteins. A high percentage of patients with biliary sludge eventually develop gallstones [19]. The fact that 11 of our patients had CBD biliary sludge supports such an approach. Recently, MRCP became a popular, valuable, and non-invasive investigation that replaced ERCP as a diagnostic procedure in many centres. MRCP can be used to select those in actual need for ERCP. This will limit the number of diagnostic ERCP and make ERCP more of a therapeutic tool. MRCP however is not readily available in every centre which limits its widespread application.

Although pediatric side-viewing duodenoscopes are now available, we used the adult size side-viewing duodenoscope without difficulty. This was done under sedation in 72.8% of our patients. General anaesthesia was used in young children to obviate ventilator compromise. Our success rate of ERCP was 96.8% which is comparable to the 93% success rate reported by Buckley and Connon and the 96% success rate reported by Allendroph et al. [8, 20]. It is now possible and as a result of advancements in pediatric duodenoscopes to perform ERCP in infants. In fact and as a result of this the indications for ERCP in the pediatric age group have been extended to include the investigation and treatment of a variety of biliary and pancreatic diseases including biliary atresia, choledochal cyst, pancreatitis, traumatic pancreatico-biliary ducts disruption and post liver transplant bile leak [7, 21–24]. Nine of our patients had pancreatitis and two of our patients had posttraumatic bile leak. In those with pancreatitis, ERCP proved useful. In three, it was normal, in three there were gallstones, in two the CBD was dilated without stones but an enlarged, inflamed papilla suggestive of recent stone passage, two had choledochal cysts and one had pancreatic cysts. In both children with posttraumatic bile leak, the bile leak stopped after endoscopic sphincterotomy.

In 1982, Cotton and Leage [25] performed the first sphincterotomy for a 16-year-old child with chronic pancreatitis. Since then, sphincterotomy has been reported with increased frequency and minimal morbidity. In 1992, Guerlrud et al advocated ERCP and sphincterotomy to treat children with CBD stones after prior cholecystectomy [9]. We found ERCP and endoscopic sphincterotomy valuable both pre and post laparoscopic sphincterotomy and since its use in our hospital, none of our patients required laparoscopic or open CBD exploration. ERCP in our settings proved useful both as a diagnostic and therapeutic investigation with no mortality and minimal morbidity. Pancreatitis, bleeding, cholangitis and retroperitoneal perforation have been reported as complications of ERCP [26, 27]. We have encountered only one case of bleeding at the time of sphincterotomy which stopped with adrenaline injection and four of our patients developed mild cases of pancreatitis.

In conclusion, ERCP in the pediatric age group is safe both as a diagnostic and therapeutic procedure. It can provide valuable information which aid in the diagnosis of biliary and pancreatic diseases in children as well as therapy with the technical feasibility of endoscopic sphincterotomy. This is specially so in the era of laparoscopic cholecystectomy, where ERCP should be the treatment of choice in children with CBD stones who are going or have previously undergone laparoscopic cholecystectomy.

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