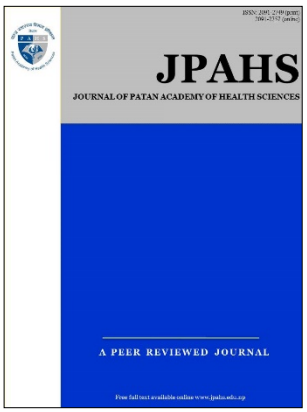

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Laparoscopic excision of congenital choledochal cyst in children and modified hepaticojejunostomy with an unequal length of the jejunal loop

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Abstract

Introduction: Complete excision biliary-enteric reconstruction is necessary for a congenital choledochal cyst (CC) to prevent recurrent cholangitis, acute pancreatitis, and cholangiocarcinoma. Among various reconstructions, this study aims to evaluate the therapeutic effect of unequal length jejunal loop for the biliary reconstruction of congenital choledochal cyst.

Method: The clinical data of 56 cases of congenital choledochal cyst treated in the pediatric surgery department of Children's Hospital Affiliated to Zheng Zhou University were retrospectively analyzed. All cases were treated with choledochal cyst resection and unequal length jejunal loop biliary reconstruction, including 51 cases with laparoscopic surgery and 5 cases with traditional surgery.

Result: Choledochal cyst resection and unequal length jejunal loop biliary reconstruction were successfully completed in all cases. One case of laparoscopic operation developed biliary fistula on the 3rd day after the operation, and the biliary fistula healed after conservative treatment for 8 days. The other cases recovered smoothly without obvious complications. No contrast agent bile loop reflux was found in upper gastrointestinal angiography.

Conclusion: The modified jejunal loop biliary reconstruction has many advantages over the traditional biliary reconstruction, which is worthy of clinical application.

Keywords: biliary enteric reconstruction, congenital choledochal cyst, hepaticojejunostomy, laparoscopy, Roux-en-Y

Introduction

A congenital choledochal cyst (CC) is a cystic dilatation of the biliary ducts, which causes recurrent cholangitis, acute pancreatitis, and cholangiocarcinoma.[1,2] Excision of a cyst and biliary-enteric reconstruction based on Todani classification[3], includes resection of the cyst and various biliary enteric reconstructions for type I and IVB CC. Open or laparoscopic Roux-en-Y hepaticojejunostomy (HD) or hepaticoduodenostomy (HD) are used, though most surgeons favor HJ for a tension-free anastomosis and to prevent reflux of the enteric contents into the biliary system which causes recurrent cholangitis and scarring. Low surgical morbidity with reliable long-term outcome in children has been reported following HJ in a multicenter study from China.[4] Open surgery for excision of CC and modified biliary-enteric anastomosis with "uncut" jejunum loop and occlusion of afferent limb together with a side-to-side jejunostomy has several benefits, for example, reduced morbidity by avoiding the possible effect on blood supply and contamination due to division of jejunum.[5] We present our single-center experience of this modified approach of HJ biliary-enteric reconstruction with unequal, uncut jejunal loop after resection of CC in children.

Method

This was a cross-sectional study from November 2017 to December 2020 at Children Hospital, Zheng Zhou University, Henan, China. Consecutive cases of children diagnosed for congenital CC who underwent authors' modification of the uncut, unequal length of jejunal loop hepaticojejunostomy biliary-enteric reconstruction were included in this study. The study was approved by the hospital ethical committee.

Preoperative clinical presentations and demographic variables were recorded for analysis. Findings of color Doppler Ultrasonography (USG) and Magnetic Resonance Cholangiopancreatography (MRCP). As per hospital protocol all children had undergone routine MRCP examination, contrast upper gastrointestinal (UGI) radiography during and/or after surgery. Surgical complications (bile leak, reflux), in-hospital mortality, and follow-up (abdomen physical exam, USG, UGI contrast radiography) data for up to 2 y were analyzed descriptively frequency and percentage. Microsoft Excel was used for analysis.

Authors' modification to Warren Roux-en-Y jejunal loop biliary-enteric reconstruction included: at a point 20 cm and 50 cm from Tretz ligament, the jejunum was folded to create a loop of 30 cm, and a side-to-side jejuno-jejunostomy (JJ) of 4 cm was created. The bowel loop was brought out via umbilical port for side-to-side jejunal anastomosis. The anastomosis was performed outside the abdominal cavity manually using 5-0 absorbable sutures. Proximal to JJ anastomosis the seromuscular layers of both limbs of the jejunum were sutured to stabilize the loop and reduce strain on JJ anastomosis. After the completion of JJ anastomosis, the jejunal loop was put back into the abdominal cavity, and pneumoperitoneum reestablished to continue the laparoscopic procedure.

The jejunal loop was pulled up to porta hepatis through transverse colon mesentery for HJ reconstruction. The HJ was performed to maintain an anastomotic diameter of 1 to 2 cm size. Proximal to HJ, at 5 cm, i.e., at a midpoint between HJ-JJ, the lumen of the afferent limb was occluded. For occlusion, a number 2 silk ligature was passed via avascular mesentery close to bowel wall and knot tied with moderate strength just to occlude the lumen and not to strangulate the intestine, Figure 1. An objective guide we follow at our hospital for the tightness of occlusion is to leave just enough space to allow the tip of a mosquito forceps to pass between the suture and the intestinal wall.

Post-operative management and regular follow-up. A per hospital protocol, follow-up included clinical abdominal examination, upper gastrointestinal contrast radiography performed between 8th to 12th day of surgery to check for reflux, color Doppler USG of liver,

pancreas, and liver function test, serum amylase levels).

The hospital stays and in-hospital mortality were recorded.

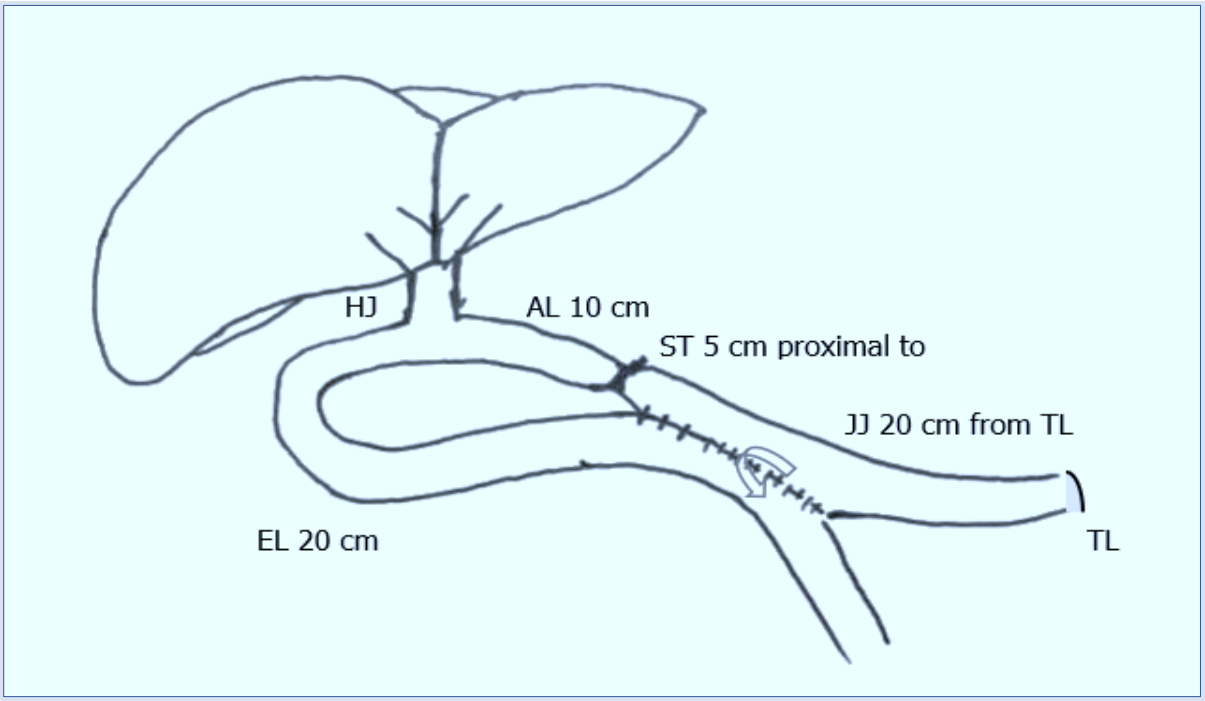

Figure 1. Laparoscopic excision of congenital choledochal cyst in children. Modified biliary-enteric reconstruction with uncut unequal jejunal loop of 30 cm, short afferent limb of 10 cm (AL), and long efferent limb of 20 cm (EL). The lumen of AL lumen occluded with silk tie (ST) 5 cm proximal to hepaticojejunostomy (HJ). A side-to-side jejuno-jejunostomy (JJ) at 20 cm from Treitz ligament (TL).

Result

Total 56 children underwent resection of congenital choledochal cyst and HJ modified biliary-enteric reconstruction using the uncut jejunal loop of unequal length comprising of short afferent and long efferent jejunal limb. Laparoscopic surgery completed in 51 and laparotomy in 5 (2 re-operation after external drainage of cyst and 3 for recurrent pancreatitis with slightly dilated common bile duct). Age ranged from 3 mo to 11 y, an average of 3.2 y; 12 under one y, 24 between 1-3 y, and 20 were 4-11 y of age.

Clinical presentations of cyst included abdominal pain in 47, jaundice in 21, white clay color stool in 9, and recurrent pancreatitis in 3 children. Fifteen children were diagnosed incidentally during abdomen USG examination. Preoperative color Doppler USG and MRCP showed Todani type-I CC in 47 children (34 cystic, and 13 fusiform dilatation), type IV in 6, and mild dilatation of common bile duct in 3 children with recurrent pancreatitis.

Intraoperative cholangiographs performed in 32 cases were consistent with preoperative cc classification. Abnormal pancreaticobiliary junction was found in 25, including 3 cases who had repeated pancreatitis with mild dilatation of the common bile duct.

One case developed biliary leakage on the third postoperative day. The fistula healed after conservative treatment for 8 d.

During follow-up, upper gastrointestinal contrast radiography performed between the 8^{th} to 12^{th} day of surgery revealed no reflux. During a maximum follow-up of 2 y and 1 mo, all children had normal findings on abdominal examination, color Doppler USG of the liver, pancreas), normal liver function test, and serum amylase levels. There was no in-hospital mortality.

Discussion

We found that laparoscopic resection of congenital choledochal cyst followed by

modified biliary-enteric hepaticojejunostomy with the uncut jejunal loop of unequal length (shorter afferent loop and longer efferent loop) was a safe, effective and simple procedure. The modification of side-to-side JJ and occlusion of the lumen of the afferent loop at 5 cm proximal to HJ successfully prevented reflux in all cases as observed during postoperative follow-up with upper gastrointestinal contrast radiography performed between 8th to 12th day of surgery. During a maximum follow-up of 2 y and 1 mo, all children had normal findings on abdominal examination, color Doppler USG of the liver, pancreas), normal liver function test, and serum amylase levels. The average postoperative hospital stay was 7 d and no in-hospital mortality in our series.

Report from the open surgical approach in patients with choledochal cyst excision with similar modified jejunal loop for HJ biliary enteric reconstruction has shown satisfactory outcome.[5] A recent systematic review shows laparoscopic approach has similar or even improved outcomes in terms of reduced hospital stay than open surgery for children.[6]

Only one of our cases developed a biliary leak on the 3rd postoperative day which healed by conservative treatment of 8 days. Biliary leakage is not a common occurrence. The overall short-term complications after the laparoscopic procedure in children are low (7.1%), with bile leakage reported in 2.8% (9/325), and intestinal necrosis in 0.3% (1/325).[7] However, in patients younger than 3 mo of age the complications are high up to 21.1% (4 cases) and thus requires extra attention.[7] Similar low recurrences of bile leak of 2% (in 8 out of 325 cases), and spontaneous closer in most of them (7 of 8) was reported, with only one case of bile leak requiring a second operation.[8]

Our modification of the uncut jejunal loop to the Warren biliary-enteric anastomosis proposed in 1965[9] has several benefits. Research has shown, that not transecting the jejunum helps preserve smooth muscle and interstitial cells of Cajal network continuity for restoration of intestinal migrating motor complex unlike the conventional Roux-en-Y procedure requiring transection of jejunum which impairs the bowel motility and affect the postsurgical recovery of bowel function after biliary-enteric reconstruction.[10,11] The uncut jejunal loop reconstruction has improved myoelectrical continuity, helps maintain peristalsis in the bile duct-enteric loop, prevents stasis of food and bile in the anastomotic limb.[12] Furthermore, the modified uncut jejunal loop is simple, saves time, avoids contamination by bowel contents, and has less risk for compromise to a blood supply by avoiding damage to the bowel mesentery.

The choice of biliary reconstruction varies with personalized methods of minimally invasive reconstruction and favors HD to reduce the operative time and postoperative morbidity.[13] Reports show that laparoscopic excision of the cyst with HD has a shorter operative time, and is not inferior to HJ.[8,14] However, a high incidence (33.3%) of complications due to reflux after HD deems it not an ideal method of biliary-enteric reconstruction.[15] Reported incidence of cholangitis during follow-up is 3 times more common in HD (1.5% of 328 HD) than HJ (0.6% of 162 HJ); and also a higher incidence of gastritis after HD (3.8%).[8]

The biliary diversion procedures have some evolutionary milestones for the establishment of Roux-en-Y in biliary enteric bypass (for hepaticojejunostomy or cholecystojejunostoy or choledochojejunostomy) as a favored procedure for effective internal biliary drainage for a tension-free biliary-enteric anastomosis.[16] A 40 cm of jejunum is considered an adequate bypass length between the biliary system and jejunum for reduction of reflux. The term hepaticojejunostomy (HJ)[17] was introduced in 1949 by Sanders, and the term Roux-en-Y (for choledochojejunostomy)[18] by Allbritten Jr. The modification by Warren in 1965 using an uncut loop of jejunum with occlusion of the proximal limb with mattress suture was introduced.[9] Same modification was proven satisfactory to prevent the reflux after gastrectomy, and clinical trials in effectively reducing the incidence of reflux.[19,20] It was confirmed in animal experiments that the pressure of the

biliary tract and the detection rate of intestinal bacteria in Warren's anastomosis were lower than those in Roux-Y anastomosis. Reflux and cholangitis are reported to occur 3 times more commonly in HD (1.5% of 328) than HJ (0.6% of 162).[8]

Laparoscopic surgery for congenital choledochal cyst became widely used after the successful case report in 1995 by Warren of cyst excision and HJ reconstruction for a girl of 6 y of age.[21] Further modification of the unequal length of the uncut jejunal loop with a shorter afferent and longer efferent limb in open surgery for choledochal cyst was reported to achieve a favorable result in reducing the incidence of stasis, torsion of the loop, and subsequent complications.[6] The longer jejunal loop has an increased risk of torsion, herniation, and necrosis. We had observed such a case at our institute after 1 y following the excision of the choledochal cyst and Warren's Roux-en-Y reconstruction for resection of CC. W later modified the technique at our hospital with HJ using the uncut jejunal loop of unequal length in a child who required laparotomy and excision of gangrenous bowel.[22]

In our study, the modified uncut jejunal loop for HJ reconstruction, the afferent loop had a length of 10 cm in comparison to the efferent limb of 20 cm. We occluded the lumen of the afferent loop by number 2 silk ligature to avoid food reflux and the tie was placed close to the HJ, at 5 cm to avoid the blind loop. The ligature was passed through an avascular area of the mesenteric and tied with moderate tightness just enough to occlude the lumen (the ligature could accommodate the tip of the hemostat) and not strangulating the bowel. The occlusion helps prevent isoperistaltic reflux of intestinal content into the biliary tree, and thus, reduces the retrograde infection and cholangitis.

On types of biliary reconstruction, the HJ-Roux-en-Y in 49 cases, HJ- modified Warren in 31 and HD 32 have shown similar surgical outcomes (P>0.05) for complications and hospital stay. But, intrahepatic reflux detected by ultrasonography during follow up was common and observed in 37.5% of HD patients.[23]

On surgical approach, laparoscopic and robotic-assisted excision of choledochal cyst has shown similar outcome to that of laparotomy. The minimally invasive approaches of laparoscopy and robotics have better cosmesis but at a higher cost and technological demands.[24]

Complete excision of the cyst, similar to the type of reconstruction, has an important role to prevent complications and malignancy. Analysis of 267 adult patients with type-I CC from 1998 to 2015, complete resection 171 vs. remnant 1-cm proximal cyst wall in 96 have shown that complications were twice as much in the remnant group (28.1% vs. 14.0%), p=0.005), especially cholangitis (7.3% vs 1.2%, p=0.021). The long-term postoperative complications of anastomotic stricture, reflux cholangitis, intrahepatic bile duct stones were not significantly different, the biliary malignancy occurred in one patient in both groups (complete resection patient at 25 mo, remnant patient at 5 mo) and one anatomical site malignancy in remnant (at 10 mo).[25] The natural history of choledochal cysts in adults treated by internal bypass drainage procedure shows the high incidence of cancer up to 30% compared to 0% after complete excision of the cyst.[26,27]

Since the end of 2017, we have modified Warren's Roux-en-Y HJ by the uncut unequal length of the jejunal loop, with a shortened loop length of 30 cm which includes a shorter 10 cm of afferent limb and longer 20 cm efferent limb. We continue to use the side-to-side JJ and occlusion of the afferent limb in-between HJ and JJ. After a follow-up of a maximum of 2 y among 56 cases, we had no obvious postoperative complications.

Conclusion

The laparoscopic approach of excision of congenital choledochal cyst in children with modified biliary reconstruction using un-cut unequal length jejunal loop with shorter proximal and longer distal limb, together with

a side-to-side jejuno-jejunostomy combined with occlusion of the proximal limb between hepaticojejunostomy and jejuno-jejunostomy is a safe technique. This modified technique of biliary-enteric bypass following complete excision of choledochal cyst in children has reduced incidence of anastomotic leakage or necrosis, avoids reflux, and the risk of torsion of biliary-enteric loop and internal hernia.

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Conflict of Interest

The authors declare that they have no competing interests

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Author Contribution

Concept and design: ALL; data collection: Liu Ru Yi and Li Peng Fei; data analysis: Hou Guang Jun; draft of the manuscript: Hou Guang Jun; Final draft review: ALL

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