Use of vascularized appendiceal graft for biliary tract replacement in a case of choledochal cyst

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ABSTRACT

A case of a choledochal cyst managed with cyst excision and an antiperistaltic hepaticoappendicoduodenostomy (HAD) is reported. Although the possibility of a postoperative evaluation using endoscopic retrograde cholangiopancreatography has been considered previously, this is probably the first reported case where postoperative stenting of HAD has been successfully performed to treat postoperative biliary leak.

KEY WORDS: Choledochal cyst, appendix, hepaticoappendicoduodenostomy

INTRODUCTION

Roux-en-Y hepaticojejunostomy (HJ) is the most frequently used biliary reconstruction procedure in the management of choledochal cysts. This is both nonanatomical and unphysiological but is also prone to complications like cholangitis. Numerous modifications of intestinal conduits to prevent reflux have been reported in biliary reconstruction, but none have proven entirely satisfactory in eliminating cholangitis. Sporadic case reports and a few series of biliary duct substitutions with the appendix have been reported. We report a case of a choledochal cyst managed with cyst excision and an antiperistaltic hepaticoappendicoduodenostomy (HAD). The use of the appendix for biliary tract replacement appears promising.

CASE REPORT

An 11 year-old girl was admitted for acute pain in the right upper abdomen. This was the first episode of pain and was also associated with fever. There was no history of vomiting or icterus. The abdominal examination was unremarkable except for tenderness in the right hypochondrium. The routine blood investigations were unremarkable except for leucocytosis. The liver and renal function tests were normal. An ultrasonography of the abdomen showed a dilated common bile duct (CBD) with a normal liver. Endoscopic retrograde cholangiopancreatography (ERCP) showed a fusiform dilatation of the common bile duct although the pancreatic duct was normal [Figure 1].

A laparotomy performed through the right subcostal incision confirmed the presence of a choledochal cyst was subsequently excised. The caecum and the ascending colon up to the hepatic flexure were mobilized so that the appendix could lie next to the duodenum. The appendix was then divided for about 4-5 cm. The caecal defect was closed in two layers. The appendiceal graft patency was confirmed.

Figure 1: Preoperative ERCP confirmed the diagnosis of fusiform type of choledochal cyst.
The wider caecal end of the appendix was now anastomosed to the common hepatic duct in an end-to-end manner using single layer 5-0 Vicryl suture. The appendiceal graft was then refashioned by excising the distal two cm to prevent kinking. The appendix was then anastomosed to the posterior aspect of the 2nd part of the duodenum in an end-to-side fashion using 5-0 Vicryl suture. No antirefluxing tunnel or serosal hitch was employed. There was a blood loss of 50 cc intraoperatively. Total duration of surgery was 1½ h.

Postoperatively, the patient developed a biliary leak for which she was referred for ERCP. Side-viewing endoscopy revealed that the neopapillary opening was placed proximally and lateral to the native papilla. Selective cannulation of the appendicular graft through the neopapilla showed a small leak at the distal end. A 7 Fr plastic biliary stent was placed across the leak with the upper end of the stent in the left hepatic duct [Figure 2].

The presence of an appendico-duodenal anastomosis at the postero-medial aspect facilitated the procedure; it would have been technically difficult, if not impossible to stent the HAD if the stoma would have been placed at the antimesenteric aspect of the 2nd part of the duodenum. The child has been found to be doing well clinically in a three-year follow-up visit. A hydroxy-iminodiacetic acid (HIDA) scan showed no evidence of any cholestasis.

**DISCUSSION**

The use of the appendix as a substitute is not a new concept. It is commonly used in creating Mitrofanoff’s continent vesicostomy as well as for the Malone antegrade continence enema (MACE) procedure in bowel incontinence. The appendix has been occasionally used as a replacement for part of the ureter and urethra and has been shown to have long-term patency and functionality.[4-6] It has also been sporadically used for biliary tract reconstruction in the last two decades.

The use of appendicular graft in biliary reconstruction was first reported in an experimental animal model by Grossfield et al.[7] Ten dogs were subjected to replacement of the biliary tract using an appendiceal graft with excellent results. The appendix was anastomosed to the duodenum in an end-to-side manner and no antirefluxing tunnel was used. Late results showed healthy patent vascularized grafts at six months to one year with no evidence of stricture, bile stasis or calculus formation. Later, Greenhoze and Lily successfully used an appendiceal r graft for independent drainage of the right anterior hepatic segment through an accessory bile duct to a Roux-en-Y hepaticoportoenterostomy.[8] Valla described the first case in which the appendix alone was used as a replacement of the bile duct in a case of biliary atresia with good postoperative results for 2 years.[9] Gupta and Rohtagi successfully used an appendiceal conduit for hepatporto-jejunosumosty (HAJ) for advanced cases of biliary atresia.[10] Subsequent reports of the use of the appendix for biliary tract reconstruction in choledochal cysts have shown promise.[3,11,12] Most of the published cases showed good immediate, early as well as late results.

The advantages of using the appendix as a replacement for the bile duct include:

1. Small caliber vascularized patent conduit anatomically similar to the native bile duct.
2. Allows drainage of bile into the duodenum as apposed to the roux loop and is more physiological.
3. Avoids a long roux loop with its attendant problems of blind loop, stasis and need for intestinal anastomosis.
4. Technically simple procedure with fewer suture lines, which can be done expeditiously.
5. Has the potential to decrease the incidence of postoperative cholangitis.[10]
6. Allows postoperative evaluation using endoscopic techniques (ERCP) - not possible in other kinds of bilioenteric derivations.[9]

A few technical issues about the procedure need to be highlighted. One relates to the length of the appendix -using a long graft invites kinking and surgical failure. An antireflux measure as previously described by some authors was not used in our case. We believe that tunneling of the appendix in a thin-walled duodenum could be technically difficult, if not impossible. One more issue that needs to be addressed is isoperistaltic...
versus antiperistaltic HAD. We tend to agree with a recent report from China that anastomosing the caecal end of the appendix to the common hepatic duct seemed more favorable than the other way around, because the caecal end could be easily trimmed to the size of the common hepatic duct, which was more or less dilated in the presence of a choledochal cyst.\textsuperscript{[11]}

A retrospective multicentric study published five years ago, however, has refuted the safety of appendix grafting for biliary reconstruction in children.\textsuperscript{[13]} Five cases of choledochal cyst treated with isoperistaltic HAD in three pediatric centers in France were reviewed. Postoperatively, all patients initially became asymptomatic but developed laboratory evidence of anicteric cholestasis within one year. The most common manifestation was an increased gamma-glutamyl-transpeptidase level, whereas histologic findings showed liver damage (mainly fibrosis).

Reoperation had been carried out in four patients; the graft procedure was converted to HJ in three and to cholecodochoduodenostomy in one. Surgical exploration showed kinking in one patient and stenosis in one. In the remaining two cases, there was no discernible cause of cholestasis and appendix histology findings were normal. In all four reoperated patients, liver function findings returned to normal within one month. The authors suggested that an appendiceal graft should be used only as a salvage technique when conventional HJ repair is contraindicated.

Due to the high risk of graft dysfunction in this series, they recommended screening tests to detect biochemical or histologic cholestasis in any patient previously treated with appendix grafting. Secondly, there had been a few reports of appendicitis complicating the use of appendiceal conduits in MACE and Mitrofanoff procedures.\textsuperscript{[14,15]} However, we believe that it is too premature to write off a procedure based on a couple of case reports. We believe that the use of the appendix for biliary tract reconstruction holds promise and it provides an ideal physiological conduit that would allow a free flow of bile from liver with minimal risk of cholangitis, stricture or stasis.

\textbf{REFERENCES}


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