



Operative Technique

Inspissated bile syndrome; Safe and effective minimally invasive treatment with percutaneous cholecystostomy in neonates and infants



Bapesh K. Bollu ^{a,*}, Michael J. Dawrant ^a, Kunal Thacker ^b, Gordon Thomas ^a, Murthy Chenapragadda ^c, Kevin Gaskin ^b, Albert Shun ^a

^a Department of Paediatric Surgery, The Children's Hospital at Westmead, Sydney, Australia

^b Department of Gastroenterology, The Children's Hospital at Westmead, Sydney, Australia

^c Department of Radiology, The Children's at Hospital Westmead, Sydney, Australia

ARTICLE INFO

Article history:

Received 9 April 2016

Received in revised form 6 September 2016

Accepted 7 September 2016

Key words:

Inspissated bile syndrome

Percutaneous cholecystostomy

ABSTRACT

Purpose: Inspissated bile syndrome (IBS) is a rare cause of obstructive jaundice in neonates and infants with several treatment options reported. We present our experience with the use of minimally invasive ultrasound-guided percutaneous cholecystostomy drain catheter placement with ongoing saline lavage in neonates and infants.

Methods: Retrospective chart review of patients treated with percutaneous cholecystostomy, from February 2010 till June 2015. We reviewed the technical and clinical success along with complications of the procedure.

Results: There were 6 patients, mean age 17 weeks (range 4–40). Most had significant risk factors for IBS presenting with biliary obstruction. A total of 7 procedures performed on the 6 patients, with a technical success rate of 6/7. One patient required cannulation of the intrahepatic biliary system. Drains were flushed for a median of 26 days (10–70). Clinical success was achieved in all patients. 3 had displacement of the drain, one of which required re-insertion. Another developed a small sub-hepatic collection post procedure with pyrexia. On long term follow up one was found to have a forme fruste choledochal cyst.

Conclusion: Centers with suitable interventional radiology services ultrasound-guided percutaneous cholecystostomy drain catheter placement with ongoing saline lavage is a safe and effective minimally invasive treatment for IBS in neonates and infants.

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1. Background/Purpose

Inspissated bile syndrome (IBS) is a rare cause of cholestatic jaundice. In one case series it accounted for around 8% of the infants with a surgical cause for their jaundice, with an estimated incidence of 1 in 175,000 live births [1]. Spontaneous resolution of this condition with or without oral ursodeoxycholic acid is well reported [2–4]. However, when obstruction persists, intervention is required to relieve the obstruction. Traditionally this has been performed as an open procedure [5,6]. Ultrasound (US)-guided percutaneous cholecystostomy drain insertion and irrigation of the biliary system provides a simple minimally invasive treatment option but there has been little published on this [7–9]. We present our experience with this technique.

2. Patients and methods

This study was approved by the Human Research Ethics Committee of the Sydney Children's Hospital Network (LNR/15/SCHN/305). A

* Corresponding author at: Department of Paediatric Surgery, Women's and Children's Hospital, North Adelaide, Adelaide, South Australia, 5006, Australia. Tel.: +61 433545060; fax: +61 8 8161 6968.

E-mail address: bapeshbollu@gmail.com (B.K. Bollu).

retrospective review of the medical records of all children below one year of age that underwent a US-guided percutaneous cholecystostomy catheter insertion and irrigation of the biliary system for IBS, at the Children's Hospital at Westmead, from January 2010 to June 2015 was done. A total of 6 patients (requiring 7 procedures) were identified from the hospital operating theater database. Patient demographics, details of the presentation, associated medical conditions and investigations were recorded. Clinical progress was documented particularly noting the technical and clinical success of the procedure along with any associated complications.

The procedures were performed under general anesthesia in our interventional radiology suite. Prophylactic antibiotics were given pre-operatively to all patients. The placement technique involved a US-guided percutaneous approach to place the catheter into the gallbladder using a trocar technique. The catheters were placed through the inferior margin of the liver adjacent to the gallbladder. In 6 out of the 7 procedures a 6.3Fr Dawson-Mueller Catheter (Cook Medical, Bloomington, IN) was used. After insertion of the catheter, contrast (Omnipaque 240) was injected to confirm the correct placement of the catheter and to perform a trans-cholecystic cholangiogram to confirm the biliary anatomy and obstruction (Fig. 1A). The gall bladder could not be accessed in one patient (patient D, Table 1), so the drain was placed into a dilated bile duct

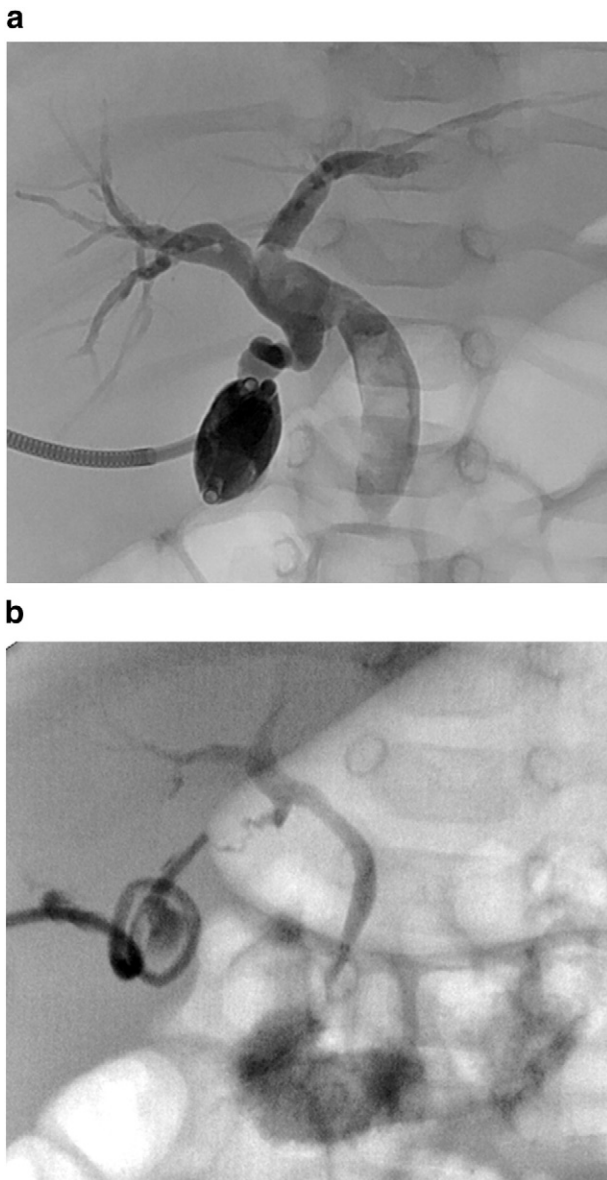


Fig. 1. A. Cholangiogram during drain placement; outflow obstruction of CBD with proximal dilatation. B. Cholangiogram just before removal of drain 3 weeks later showing decompressed biliary system with rapid flow into the duodenum.

through the left lobe of the liver. In another patient (patient E, [Table 1](#)), 4Fr Skater™ Centesis Catheter (Argon Medical Devices, Athens, TX) was used, however as it had been ineffective and had become displaced, the drain was replaced with a 6.3Fr Dawson-Mueller Catheter after 7 days. The drains were left on free drainage initially and then started on normal saline flushes twice daily (3–5 ml). Patient follow up to assess clinical improvement was done via clinical assessment, biochemical markers and a repeat cholangiogram to confirm resolution of obstruction within the biliary system ([Fig. 1B](#)).

3. Results

There were 6 patients (5 males), mean age of 17 weeks (range 4–40). Basic demographics of the six patients, their past medical history and presentation are outlined for each patient in [Table 1](#). Commonly recognized risk factors were present in 5 of the 6 patients.

The diagnosis of IBS was made with an abdominal US, as part of the first-line investigation of cholestatic jaundice in each of these patients. All of the cases showed a degree of intra- and extra-hepatic duct

dilatation with some sludge-like material in the biliary system. The median extra-hepatic bile duct dilatation was 5.1 mm (range 2.3 mm–5.8 mm). The child with only mild extra-hepatic duct dilatation (child B- common bile duct 2.3 mm) had acoustic shadowing of the biliary sludge suggesting that they might also have fully formed stones. This child had a normal bilirubin level but the rest of the liver function tests were deranged: ALT 469 umol/dL, AST 166 umol/dL and GGT 165 umol/dL. The patients were all placed on oral ursodeoxycholic acid at 15 mg/kg twice daily. It was continued for a median of 5 days (4–7) prior to progressing to intervention. All of the patients had a DISIDA (technetium labeled diisopropyl iminodiacetic acid) scan, around 1 to 3 days after their initial ultrasound, as part of their work up to assess for biliary obstruction. The nuclear medicine physicians reported satisfactory parenchymal function of the liver with abnormal excretion in all of the patients; 5 patients had no excretion of bile into the gastrointestinal tract whilst patient B showed partial excretion. All of these patients had no clinical or biochemical improvement in their conditions, so the decision was made to progress to US-guided percutaneous cholecystostomy catheter placement with ongoing saline lavage.

Technical success was defined as being able to successfully place the percutaneous cholecystostomy catheter. A total of 7 procedures were performed on the 6 patients. 6 of the 7 procedures were done without complication and were deemed to be technically successful. However for patient D the gallbladder was not able to be accessed, so using a trans-hepatic approach the catheter was placed at a dilated portion of the confluence of the common hepatic ducts. Patient E required 2 procedures. Initially a 4Fr Skater™ Centesis Catheter was used, however as it had been ineffective and had become displaced, the drain was replaced with a 6.3Fr Dawson-Mueller Catheter after 7 days.

Clinical success was seen as a resolution of the patient's clinical symptoms along with the improvement of liver function tests. A cholangiogram was also done prior to removal of the catheter to confirm that the bile ducts were clear of any obstruction. All 6 patients achieved this. The catheters were left in for a median of 26 days (range 10–70). 3 of the patients could be discharged home with on-going flushes (A, C, E). The other 3 patients were long term in-patients requiring other care. Patient B had the longest duration of saline flushes. Of note this was the patient who had gallstones within the distal common bile duct along with biliary sludge.

The complications from our series were minor in nature. The most common issue was drain displacement out of the gall bladder (3 patients). In one patient, (patient E), their drain (4Fr Skater™ Centesis Catheter) had become displaced, but as the drain had been ineffective it was upgraded to the standard 6.3Fr Dawson-Mueller Catheter after 7 days. For the other two of these patients, this occurred once the gallbladder had collapsed into its normal size and they had clinically improved. The most notable complication was seen in patient D who developed some pyrexia following the procedure and was found to have a small intra-abdominal collection. This collection was presumed to be infected and the patient responded to a short course of intravenous antibiotics.

The patients have been followed up for a median of 10 months (range of 3 - 60 months) with repeat imaging and bloods tests. They remain clinically well with normal liver function tests. No recurrences of symptoms have been reported. One patient (patient C) was noted to have persistent extra-hepatic bile duct dilatation and with further investigations was diagnosed and operated on for a forme fruste choledochal cyst with a long common biliary-pancreatic channel. At initial presentation the common bile duct was 3.5 mm in diameter.

4. Discussion

In 1935, Dr. William Ladd reported inspissated bile as a cause of cholestatic jaundice in infancy [5]. He observed cases where he proposed that inspissated bile was plugging and obstructing stenotic common bile ducts. Although biliary sludge can be associated with choledochal malformations [10], in the majority of cases of IBS the patient will

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