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Spontaneous bile duct perforation in a newborn

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1. Introduction

Spontaneous perforation of the bile duct in infancy is a rare event with a reported incidence of 1.5 in 1,000,000 children and with a median age at diagnosis of 2.4 months [1]. Usually the cause is unknown, but a wide range of causes have been speculated, including delivery trauma [2]. The condition is usually managed by surgical drainage but strategies range from non-surgical treatment to biliary-enterostomy [1–3].

In the following we describe a case of spontaneous bile duct perforation in a 3-week old male, who had suffered a complicated delivery.

2. Case report

The child was born at gestational age 41 + 5 via cup-assisted vaginal birth complicated by shoulder dystocia. An Apgar score of 10 was not reached until 25 min postpartum and pH was initially 7.09. One hour postpartum pH was 7.2 with a serum-lactate concentration of 8.2 mmol/l (reference <2.0 mmol/l). The child was intubated immediately postpartum. He was treated with CPAP for 15 h and intravenous ampicillin and gentamycin for 3 days. The child was discharged 11 days post-partum, where breastfeeding was well established.

The child was readmitted 20 days old, presenting with failure to thrive, recurrent regurgitations and increasing abdominal distension but with normal bowel function. Physical examination showed a distended abdomen with peritoneal redundancy and silent abdomen by auscultation. The blood pressure was normal. Lactate concentration in peripheral venous blood was 2.5 mmol/l (reference: <2.0 mmol/l) and C-reactive protein 44 units (reference: <10 units). Abdominal ultrasonography showed no abnormalities and plain abdominal X-ray no air in the lower part of the abdomen.

At laparotomy a diffuse peritonitis with bile-stained peritoneal fluid was found.

Biliary leakage was found in the angle between the cystic duct and the common bile duct through a 1–2 mm large perforation. An intraoperative cholangiography through the opening (using a 5 French baby feeding tube) showed a normal intrahepatic biliary tree and a normally calibrated common bile duct with free passage of contrast to the duodenum but also a small amount of debris. No contrast over flow to the pancreatic duct was observed (Fig. 1). After cholecystectomy the perforation was closed using Monocryl® (Ethicon, US) 4-0 suture and the subhepatic area was drained. Postoperative treatment with intravenous Meropenem® (Fresenius-Kabi, Germany) and Metronidazole (Baxter, US) was continued for 3 days. The drain was removed on the third postoperative day. The pathologist report showed mildly inflamed gallbladder with fibrin deposition and fibrosis. Three weeks later the boy was reoperated for bowel obstruction due to peritoneal adhesions and no other pathologies were found. At 6 months follow-up the patient was wellbeing and with normal growth.

3. Discussion

A wide range of causes for spontaneous perforation of the common bile duct have been speculated, including congenital anomalies, viral infection of the common bile duct and delivery trauma [2].

In the present case the cause for perforation was obscure. However, the findings of debris in the common bile duct on the cholangiogram and the inflammation of the gallbladder makes previous obstruction of the bile duct by inspissated bile or even a passed stone a plausible cause for perforation. Since no retrograde flow of contrast into the pancreatic duct was seen, papillary obstruction seems unlikely.

The reported median age at perforation is 2.4 months with an interquartile range of 1.5–10 months, making our patient at 21 days old one of the youngest cases reported. Since our patient suffered a traumatic delivery with obstructed labour due to shoulder dystocia.
and cup-assisted delivery, trauma due to compression or traction or even ischemia due to asphyxia could also be the cause.

Management strategies range from no surgery to primary biliary-enteric anastomosis. Conservative treatment with intravenous antibiotics has also been successful [4].

Surgical drainage seems to be the preferred procedure, performed in over half of cases and with reasonable results, but 15% of patients do require a subsequent biliary-enteric anastomotic operation, primarily due to stenosis of the common bile duct [1].

Until Jeanty et al.’s review in 2015 outcomes were less clear. They found a significantly higher risk of re-operation in cases managed by surgical drainage alone (21%) compared to surgical repair of the perforation (11%). However, the total number of complications were similar in the two groups, 23% vs. 22% respectively. The most frequent complication described in both groups was bile fistula occurring in 12% of patients who had surgical drainage and 11% of patients who had primary surgical repair of the lesion.

A perforation at the junction of the cystic duct and the common bile duct is the most common site of perforation reported in 43% of described cases. Secondly the perforation is located on the common bile duct itself in 23% of cases and thirdly the perforation is found on the gallbladder in 12% of the cases [1]. Different locations of the perforation might support different management strategies. We chose to manage the perforation with cholecystectomy and primary surgical repair.

Primary suturing of the lesion was the surgical treatment of choice in 19% of the cases described, while initial cholecystectomy was only performed in 3% of the cases [1]. Suturing of a perforation at the junction site might increase the risk of subsequent stenosis of the cystic duct and hence increase the risk of cholecystitis supporting the choice of primary cholecystectomy. Re-perforations are not described in the literature, prompting early removal of external drains. Re-operations for bowel-obstruction due to adhesions is very common in patients following cholasos but seems to be rare in this population, only described in 3 out of 70 cases [1].

4. Conclusion

Our case report supports previous obstruction of the common bile duct as a possible cause for spontaneous biliary duct perforation, although delivery trauma or neonatal asphyxia cannot be excluded. Suturing of the lesion with cholecystectomy was performed. The only complication in this case was bowel obstruction due to peritoneal adhesions, which seems to be an otherwise rare complication.

Conflict of interest

The authors have no conflict of interests.

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