

Can PTCC Reduce Unnecessary Intraoperative Cholangiography for Infants' Biliary Stenosis with Cholestasis?

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Abstract: Cholestatic jaundice is a common finding in the newborn and young infants with the majority of this cases due to hyperbilirubinemia caused by multiplied conjugated bilirubin. These cases common caused by biliary obstruction due to biliary atresia or other cases. Biliary atresia happen in between 1/10,000 to 1/16,700 live births and this incidence has been consistent over time with a slightly higher on female babies. Intraoperative cholangiogram is the gold standard for diagnosing biliary atresia, but requires surgical intervention. Percutaneous transhepatic cholecysto-cholangiography are less invasive and effectively exclude biliary atresia.

Keywords: PTCC; Biliary atresia; Biliary stenosis; direct bilirubin; indirect bilirubin.

INTRODUCTION

Jaundice, is a yellowish color on the sclera, skin, mucous, and body fluids (Feldman, A.G. *et al.*, 2013). Jaundice is a common in the newborn and young infants with the majority of this cases due to hyperbilirubinemia caused by multiplied unconjugated bilirubin that will be resolve spontaneously (Cowles, R.A, 2012). In another rare case, jaundice will persist, pathological jaundice beyond 2 weeks of life. Neonatal jaundice at 2 to 3 weeks old should have fractionated unconjugated and conjugated bilirubin, which is conjugated hyperbilirubinemia is always pathologic. Persistent jaundice is abnormal and can be the a sign of hepatobiliary or metabolic problem. When jaundice continue through 2 weeks, it can be cholestasis or conjugated hyperbilirubinemia as differential diagnosis (Cowles, R.A, 2012). Sign to cholestasis include diarrhea dan poor weight gain, hypopigmented stools, dark urine, and hepatomegaly. Cholestatic jaundice affect approximately 1/2,500 infants. The number of typical disorders presenting in the neonatal period with cholestasis may be greater than at any other time in life, and may include infections, anatomical abnormalities of the biliary system, endocrine disorders, genetic disorders, and metabolic disorders, exposure to toxins and drugs, vascular abnormalities, neoplastic processes and a variety of other causes. Of the many disorders that cause neonatal cholestasis, the most commonly identifiable are biliary atresia (BA) (25–35%), genetic disorders (25%), metabolic disorders (20%), and a₁-antitrypsin (A₁AT) deficiency (10%) (Feldman, A.G . *et al.*, 2013). These cases common caused by biliary obstruction due to

biliary atresia or other cases. Biliary atresia happen in between 1/10,000 to 1/16,700 live births and this incidence has been consistent over time with a slightly higher on female babies (Cowles, R.A, 2012). Intraoperative cholangiogram is the gold standard for diagnosing biliary atresia, but requires surgical intervention (Suherly, L . *et al.*, 2022- Jensen, M.K. *et al.*, 2012). Percutaneous transhepatic cholecysto-cholangiography are less invasive⁵ and potentially effective to exclude biliary atresia (Sujka, J . *et al.*, 2018). This research aim to analyze the effectiveness of PTCC to excluded biliary stenosis from biliary atresia in infant with cholestatic jaundice to prevents unnecessary intraoperative cholangiography.

MATERIALS AND METHOD

This study is a serial cases of 5 infants with cholestasis jaundice and the laboratory result persistent direct hyperbilirubinemia. All infants underwent laboratory testing, followed by a PTCC procedures. Then compared with the laboratory of total bilirubin, direct bilirubin, indirect bilirubin and Gamma-glutamyl transferase (Gamma-GT) before and after procedures.

RESULTS

Percutaneous transhepatic cholecysto-cholangiography (PTCC) was carried out in 5 cases of neonatal, four were male and one were female in the age range of 2 months- 12 months. Then the patient underwent PTCC according to the procedure and the results were obtained biliary stenosis in 4 patients and biliary atresia 1 patients. Four patients with biliary stenosis divided to 3 diagnose, 2 patients common hepatic duct (CHD)

stenosis, 1 patient vater ampoule stenosis, and 1 patient common bile duct (CBD) stenosis. In one biliary atresia patient was kasai IIB classification of biliary atresia. Bilirubin direct, bilirubin indirek, Gamma-GT on biliary stenosis has decreased

direct bilirubin, indirect bilirubin, and Gamma-GT level. Meanwhile, on biliary atresia, there was a decrease in both total bilirubin, direct bilirubin, and indirect bilirubin, except for Gamma-GT has increased.

Table 1: Patients data

Case	1	2	3	4	5
Gender	Male	Female	Male	Male	Male
Age	4-months	2-months	12-months	3-months	3-months
Comorbidities	-	-	-	-	-
Bilirubin total					
Pre-PTCC	3.49	12.53	7.56	5.59	9.52
Post-PTCC	0.21	12.58	6.72	0.65	5.74
ΔTotal Bilirubin	↓	↑	↓	↓	↓
Direct bilirubin					
Pre-PTCC	2.97	10.25	6.70	5.31	7.59
Post-PTCC	0.11	9.76	5.07	0.36	4.96
ΔDirect bilirubin	↓	↓	↓	↓	↓
Indirect bilirubin					
Pre-PTCC	0.52	4.99	0.33	0.28	0.21
Post-PTCC	0.10	2.82	0.31	0.29	0.23
ΔIndirect bilirubin	↓	↓	↓	↑	↑
Gamma-GT					
Pre-PTCC	242	958	388	158	437
Post-PTCC	132	630	289	31	453
ΔGamma-GT	↓	↓	↓	↓	↑
PTCC Result	CHD stenosis	Wirsung duct partial stenosis	Vater ampullary stenosis	CHD stenosis	Biliary atresia

All patient data is presented in Table 1. All patients had PTCC to examination what diagnose is it. First patient baby boy 4-months underwent PTCC that show the contrast appears to fill the gall bladder. Cystic duct, right and left hepatic duct and CBD to spill into the duodenum. Distal common hepatic duct stenosis is showed. After flushing with 20 cc of NaCl and then duct is flowing. Ultrasonography (USG) shows stenosis CHD (Figure 1.). Laboratory result decreased total bilirubin 3.49 to 0.21, 2.97 to 0.11 direct bilirubin, decreased indirect bilirubin 0.52 to 0.10, and decreased Gamma-GT from 242 to 132. Second girls baby 2-months underwent PTCC that show partial stenosis on duct of wirsung, NaCl 0.9% flushing expedite the duct. Ultrasonography (USG) shows partial stenosis in wirsung duct (Figure 2.). Laboratory result increased total bilirubin 12.53 to 12.58, decrease 10.25 to 9.76 direct bilirubin, decreased indirect bilirubin 4.99 to 2.82, and decreased Gamma-GT from 958 to 630. Third patient 12 months boy baby PTCC shows the

contrast fill gallbladder, cystic duct, common hepatic duct, chole duct, and duodenum. Shows stenosis on vater ampoule at contrast injection (Figure 3.). Laboratory result decreased total bilirubin 7.56 to 6.72, decrease 6.70 to 5.07 direct bilirubin, decreased indirect bilirubin 0.33 to 0.31, and decreased Gamma-GT from 388 to 289. Forth, 3-month boy baby, PTCC show stenosis on common hepatic duct (Figure 4.) . Laboratory result decreased total bilirubin 5.59 to 0.65, decrease 5.31 to 0.36 direct bilirubin, increased indirect bilirubin 0.28 to 0.29, and decreased Gamma-GT from 158 to 31. And fifth patient was boy 3-months PTCC shows contrast filled the gallbladder. However, the contrast did not fill the common hepatic duct, cystic duct, and common bile duct that shows biliary atresia (Figure 5.). Laboratory result decreased total bilirubin 9.52 to 5.74, decrease 7.59 to 4.96 direct bilirubin, increased indirect bilirubin 0.21 to 0.23, and increased Gamma-GT from 437 to 453. In our case all patients with biliary stenosis has decrease

Gamma-GT level, but on biliary atresia patient has increase Gamma-GT.

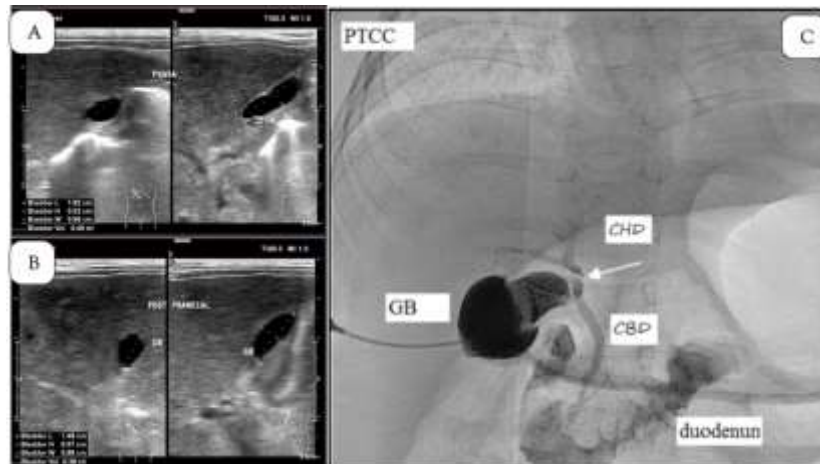


Figure 1: (A) and (B) 2-phase abdominal ultrasound, pre-prandial and post-prandial showed abnormality of contractility index gallbladder. (C) PTCC showed common hepatic duct stenosis.

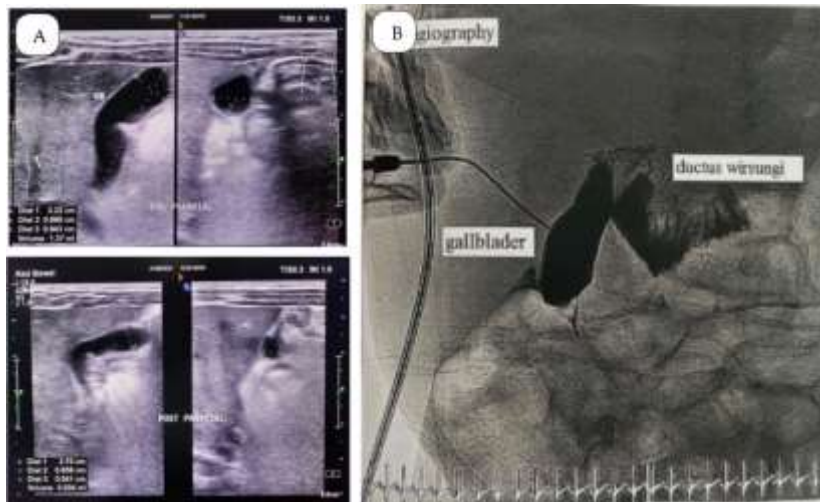


Figure 2: (A) 2-phase abdominal ultrasound, pre-prandial and post-prandial showed abnormality of contractility index gallbladder (B) PTCC showed partial stenosis in wirsung duct.



Figure 3: (A) 2-phase abdominal ultrasound, pre-prandial and post-prandial showed abnormality of contractility index gallbladder (B) PTCC showed contrast fills gallbladder, cystic duct, common hepatic duct, chole duct, dan duodenum. PTCC shows vater ampoule stenosis when contrast injection.

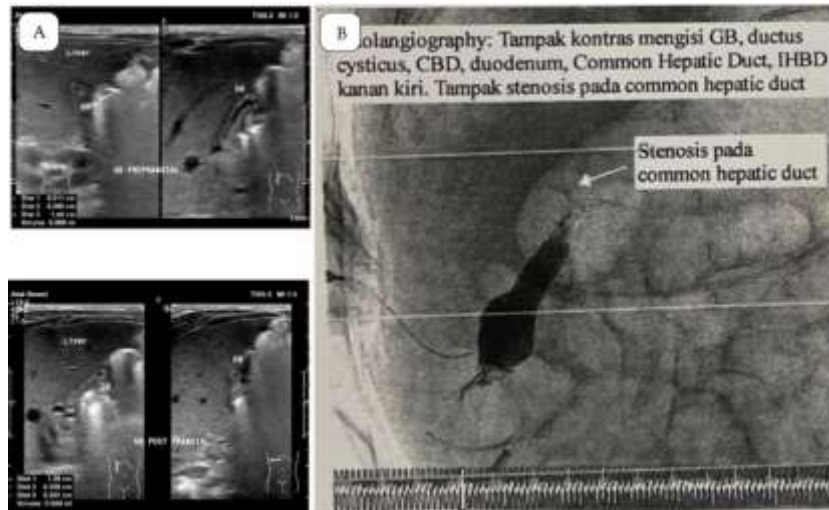


Figure 4: (A) 2-phase abdominal ultrasound, pre-prandial and post-prandial showed abnormality of contractility index gallbladder (B) PTCC showed contrast fills gallbladder, cystic duct, common hepatic duct resistance when contrast injection, then the contrast fills common bile duct and duodenum. PTCC shows common hepatic duct stenosis.

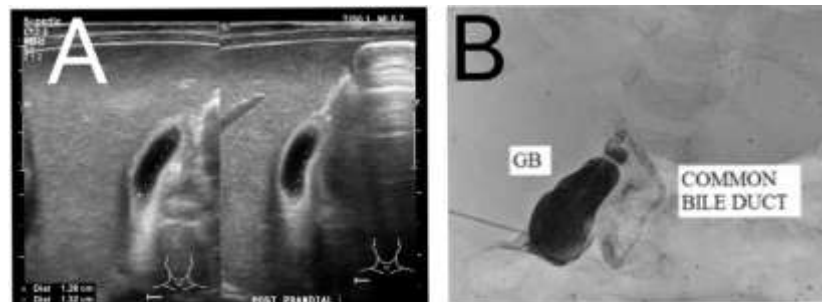


Figure 5: (A) Abdominal Ultrasound showed Abnormality of contractility index gallbladder (B) PTCC showed contrast fills gallbladder, common hepatic duct, and CBD to ampulla of Vater and spills into duodenum, contrast does not fill Bilateral IHBD and pancreatic duct (KASAI Classification IIA).

DISCUSSION

Gamma glutamyl transferase (Gamma-GT) is a plasma membrane enzyme that expressed the most in the kidney and then liver. Liver disease can increase Gamma-GT as a marker of oxidative stress and cholestasis. Liver Gamma GT found on the capillary side of hepatocytes and on the membrane of bile duct epithelial cells. Hypersynthesis of the liver or obstruction bile secretion, injury, or hyperplasia of bile duct epithelium can elevate serum Gamma-GT (Xing, M. *et al.*, 2022). PTCC or injection of contrast maybe can induce obstruction or hyperplasia respon and increase serum Gamma-GT on biliary atresia. Gamma-GT level can identify early biliary obstruction (Mysore, K.R. *et al.*, 2019). However, further research with more research subjects is needed to be able to conclude statistically the potential of Gamma-GT analysis before and after underwent PTCC.

CONCLUSION

Percutaneous transhepatic cholecystocholangiography is useful in to excluded the structure of the hepatobiliary system and to diagnose of biliary atresia in infant with cholestatic jaundice. With analyze after procedures level of direct bilirubin, indirect bilirubin and Gamma GT on stenosis biliar patient so that can prevents unnecessary intraoperative cholangiography. Differences in the laboratory of patients with biliary atresia and biliary stenosis can be seen in differences in gamma GT examination, so that the gamma GT laboratory after PTCC has increased can be a potential marker in excluding the diagnosis of biliary stenosis.

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