

CASE REPORT

Secondary infection of amoeba and typhoid fever in severe malnourished pediatric patient with Type I choledocal cyst: a case report

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Abstract

Amebiasis is caused by the protozoan *Entamoeba histolytica*. Extraintestinal amebiasis manifestation includes liver abscess and other rare manifestations involving the lungs, heart and brain. Liver abscess is the most common extraintestinal manifestation. Only few cases of amebic splenic abscess and two cases of gall bladder abscess have been reported. Typhoid fever is an infection caused by *Salmonella typhi*. It can cause extraintestinal complications such as myocarditis, endocarditis, pneumonia, empyema, osteomyelitis, arthritis, cholecystitis, spleen abscess and liver abscess. Choledocal cyst is a congenital dilation of the biliary branch. If left untreated, it can cause morbidity from cholangitis, cyst perforation, liver failure and malignancy. Until now, there is no publication about double infection of amebic and salmonella infection in a child with choledocal cyst.

INTRODUCTION

Amebiasis is caused by the protozoan *Entamoeba histolytica*. Although often asymptomatic, it can manifest as dysentery and extraintestinal amebiasis, such as liver abscess and other rare manifestations involving the lungs, heart and brain. Liver abscess is the most common extraintestinal manifestation [1]. The risk factors for liver abscess in amebiasis are still unknown [2]. There are also two case reports of amebic abscess in the gallbladder [3].

Typhoid fever is an infection caused by *Salmonella typhi*, which is a gram-negative, motile, facultative anaerobic rod. It can cause extraintestinal complications such as myocarditis, endocardi-

tis, pneumonia, empyema, osteomyelitis, arthritis, cholecystitis, spleen abscess and liver abscess [4].

Choledocal cyst is a congenital dilation of the biliary branch [5, 6]. It usually presents as a not tender right upper quadrant abdominal mass. If left untreated, it can cause morbidity due to recurrent cholangitis, cyst perforation, liver failure and malignancy [5].

CASE REPORT

A 2 years 5 months old boy with severe malnutrition came with complaints of abdominal enlargement since 6 months before admission, which was felt increasingly burdensome since

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Figure 1: Patient profile.

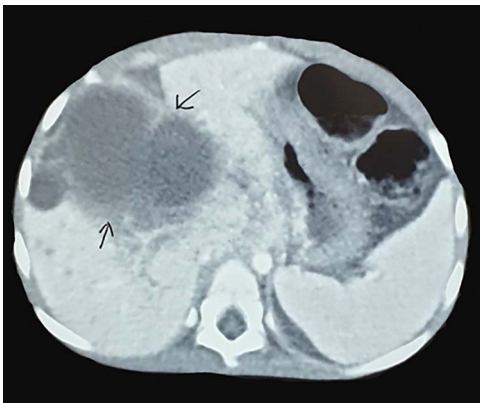


Figure 2: Abdominal CT scan with contrast.

1 month before admission. It was accompanied by low-grade fever and diarrhea > 10 times a day (Fig. 1).

Anthropometrics examination showed severe malnutrition with a body weight of 8.7 kg, body height was 87 cm and mid upper arm circumference was 9.5 cm. The result of weight for the age was -3.29 standard deviation (SD), height for the age was -0.83 SD, weight for the height was -3 SD and mid upper arm circumference for the age was -3 SD (World Health Organization growth chart). Body weight before having these symptoms was 10 kg and he was having meals three times a day with carbohydrate (rice), protein (eggs, fish and chicken), and vegetables.

Laboratory examinations showed anemia (hemoglobin: 6.3 g/dl; N: 11.5–13.5 g/dl), leucocytosis (white blood cells: 19 010/ μ l; N: 5500–15 500/ μ l), hypoalbuminemia (serum albumin: 2.03 g/dl;



Figure 3: Cyst fluid sample.

N: 3.4–5.0 g/dl), and elevated C-reactive protein (CRP) (serum CRP: 10.18 mg/dl; N: <0.3 mg/dl). Stool examination showed that diarrhea was a dysenteriform (leucocyte: 6–9/hpf). On the 15th day, IgM anti salmonella typhi (Tubex[®] TF) performed with a result of 6.00 (reactive). Blood culture revealed *Salmonella* spp. that is sensitive to ceftriaxone, piperacillin/tazobactam, cefepime, chloramphenicol, cotrimoxazole, tigecycline, aztreonam, meropenem and resistant to ampicillin/sulbactam, gentamicin, amikacin, ampicillin and cefazolin. On the 26th day, seramoeba test (Entamoebalisa IgG) performed with non-reactive result.

Abdominal ultrasound showed multiple liver cyst at the right liver lobe. Abdominal computed tomography (CT) scan revealed multiple inhomogen hypodense lesions at the IV, V, VI and VII sections of the right liver lobe, the largest lesion was 9.5 cm \times 8.8 cm \times 11.5 cm, suggestive of multiple liver cyst. Due to prolonged fever, abdominal distention and multiple cysts findings in the liver, the surgery was planned to remove the cysts that might be infected but delayed because of prolonged fever, waiting for preoperative imaging and then due to limited facility availability for postoperative intensive care unit.

The patient underwent exploratory laparotomy on the 42nd day of hospitalization. We found a dilated common bile duct adhered to the inferior side of the liver, gall bladder, transverse colon and stomach, containing 500 cc of bile fluid. No intrahepatic cyst was found. Intraoperative cholangiography revealed only extrahepatic bile duct dilatation without intrahepatic duct involvement. We performed choledocal cyst excision and Roux-en-Y hepaticojejunostomy. Amebic cysts and rod shape bacteria were found in the microscopic examination of bile fluid (Fig. 4).

The patient was diagnosed with type 1 choledocal cyst with secondary infections of amebiasis and *Salmonella typhi* with severe malnutrition. During hospitalization, the patient was given ampicillin and gentamicin for the first 3 days, and then changed based on blood culture results and clinical signs of intestinal infection to chloramphenicol and metronidazole for 40 days. The patient improved after 13 days of antibiotic and antiparasitic administration. For calory requirement we used a formula of resting energy expenditure multiply stress factor with a result 700 kcal/day consisting of 3 meals. The patient recovered uneventfully and was discharged on postoperative

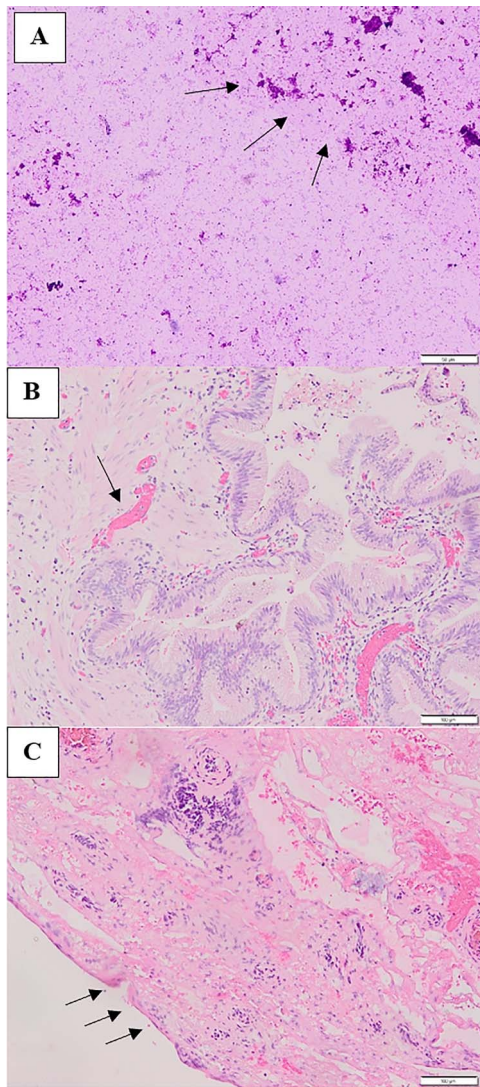


Figure 4: Histologic section of choledocal cyst and cyst fluid show (A) necrotic mass (B) erythrocyte and (C) rod shape bacteria.

Day 1. Clinical condition improved with significant weight gain after 1 month of discharge.

DISCUSSION

Choledocal cyst is a congenital dilatation of biliary tree and is most commonly found in children, especially in Asia. Pancreato-biliary malunion was thought to cause the disease. Large cysts can cause spontaneous bleeding, infection, and compression to biliary duct branches [10]. In this case, we found type I choledocal cyst, which was a dilatation of the extrahepatic bile duct. It is the most common type and seen in 75–85% of cases. Magnetic resonance cholangiopancreatography (MRCP) is the diagnostic method of choice for biliary ductal pathology. Obstruction usually due to stenotic distal duct and sludge may cause cholangitis. To prevent cholangitis and malignant progression, the definitive treatment of choledocal cyst is total cystectomy and Roux-en-Y hepaticojejunostomy.

In 1968, Babbitt's described an abnormality of pancreatico-biliary union (APBDU) in three children with choledocal cysts and hypothesized this abnormality was the cause of choledocal

cysts. APBDU is defined as the union of the pancreatic and biliary ducts outside the duodenal wall and proximal to the Ampulla of Vater. Choledocal cysts are divided into Type IA, IB, IC, II, III, IV and V [5].

Acute suppurative cholangitis is a major cause of morbidity and mortality in patients with biliary diseases. Bacteria, usually from the gastrointestinal tract through the lymphatic system or blood vessels, ascend along the bile duct and cause of inflammation of it [7]. The most common micro-organisms associated with cholangitis were *Escherichia coli* (17.5%), *Klebsiella* spp. (15.7%), *Pseudomonas* spp. (14.6%) and *Salmonella* spp. (5.8%), *S. typhi* (53.8%), *S. enteritidis* (17.9%) and *S. typhimurium* (10.7%) [7, 8, 9].

The mechanism of the presence of cystic form *E. histolytica* in bile fluid without evidence of a liver abscess is still unknown. Routes to the gall bladder could through the sphincter of Oddi that usually seen in patients who have undergone surgical intervention or could migrate to the liver via descending or hematogenous route and secreted in bile fluid [8]. The stool examination and seroameba shown negative results, this might be caused by the low sensitivity and specificity of conventional microscopy on a single stool specimen for different species of *Entamoeba*, or can also be caused by chronic amebic infections. Moreover, *E. histolytica* and *E. dispar* are morphologically identical but the last one can cause negative seroameba examination. Species should be differentiated using zymodane analysis that unfortunately we did not perform.

In this patient, *Salmonellas* spp. was isolated from both blood and stool. We believe that this patient might have had a chronic amebiasis or maybe infection by *E. dispar* and *Salmonella typhi* infection at the beginning. Secondary bacteremia supervened and severe malnourishment might have contributed to this condition because as we know, it can impair the immune system, besides the condition itself contributed to the low intake that could make the malnourished condition become severe.

CONCLUSION

Choledocal cysts should be considered in pediatric patients with infection and multiple hepatic cyst. MRCP should be done to evaluate this congenital biliary dilatation. Percutaneous cyst drainage may be needed to relieve the cholangitis faster before performing definitive surgery that might help to shorten the duration of antibiotic given. Recognition of the clinical manifestations of choledocal cysts is needed for the early management and to prevent secondary infections and complications such as further malnourishment that can affect the prognosis and recovery phase for the patient to undergo surgery.

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CONFLICT OF INTEREST STATEMENT

None Declared.

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ETHICAL APPROVAL

No Ethical approval is required.

CONSENT

Patient consent was obtained.

GUARANTOR

Dr. Riyadi adrizain.

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