



Case Report

J. Exp. Clin. Med., 2016; 33(1): 37-40
doi: 10.5835/jecm.omu.33.01.011



ERCP treatment for intrabiliary rupture of hepatic hydatid cyst in a child

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ARTICLE INFO

ABSTRACT

Article History

Received 08 / 10 / 2015

Accepted 18 / 10 / 2015

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Keywords:

Children
Common bile duct
Echinococcus granulosus
ERCP
Hydatid cyst

In this report, we present a patient with intrabiliary rupture of a hydatid cyst, which caused choledochal obstruction and treated with endoscopic retrograde cholangiopancreatography (ERCP). It is an emergency and treated with minimally invasive surgery. A twelve-year-old female patient with acute upper abdominal pain, nausea, and vomiting was admitted to pediatric surgery clinic with the preliminary diagnosis of cholestasis. There were localized right upper abdominal rebound and tenderness. Contrast-enhanced axial computed tomography images through the liver showed that there was a cystic lesion in segment 4 communicating with the biliary system. The main hepatic duct was obliterated and dilated by curvilinear densities consistent with germinative membranes expelled from the cyst into the biliary system. ERCP and sphincterotomy were performed. Germinative membrane and daughter cysts were cleaned with the use of balloon and basket. The patient is doing well without any other complication 20 months after ERCP. There have been few case reports documenting the successful use of ERCP treatment of hydatid cyst. ERCP may be a useful procedure in selected children with suspected intrabiliary rupture or common bile duct obstruction, which occurs due to daughter cyst and germinative membrane.

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1. Introduction

Hydatid cyst is endemic in Turkey, especially in the regions of Middle Anatolia, Marmara, and Thrace and causes serious health problems (Akbulut et al., 2010; Mandelia et al., 2012). Although it can develop anywhere in the body the most frequent sites are liver (60-70%) and lung (20-30%) (Mandelia et al., 2012). Here we present a twelve years old patient with intrabiliary rupture of a hydatid cyst, which caused choledochal obstruction and treated with endoscopic retrograde cholangiopancreatography (ERCP).

2. Case report

Written informed consent was obtained from patient's parent. Twelve-year-old female patient with acute upper abdominal pain, nausea, and vomiting was admitted to pediatric surgery clinic with the preliminary diagnosis of cholestasis. There were localized right upper abdominal rebound and tenderness. Abdominal ultrasound revealed a cystic mass in the liver and dilated choledochus. Laboratory results were as follows: Leukocyte count; 7.6 U/L, C-reactive protein: 1.15 mg/dL, aspartate aminotransferase: 484.2 U/L,

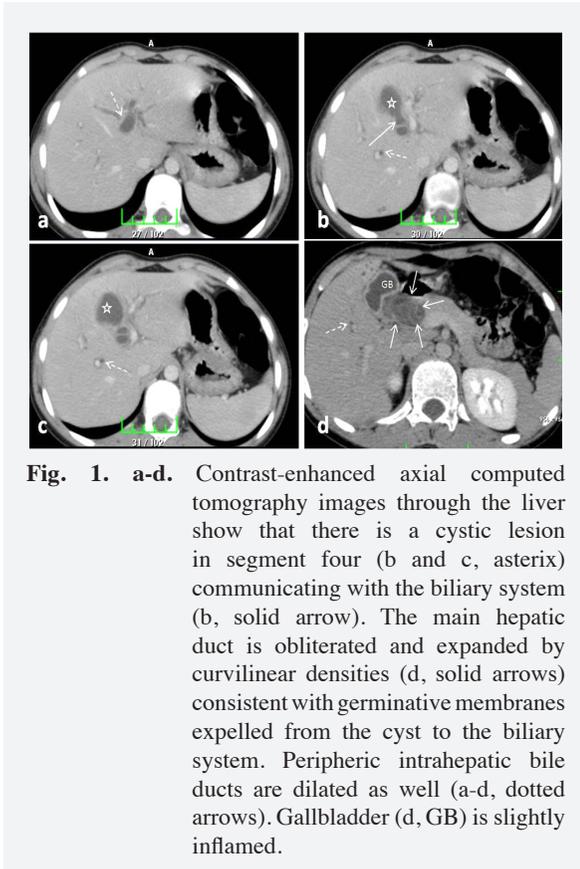


Fig. 1. a-d. Contrast-enhanced axial computed tomography images through the liver show that there is a cystic lesion in segment four (b and c, asterix) communicating with the biliary system (b, solid arrow). The main hepatic duct is obliterated and expanded by curvilinear densities (d, solid arrows) consistent with germinative membranes expelled from the cyst to the biliary system. Peripheral intrahepatic bile ducts are dilated as well (a-d, dotted arrows). Gallbladder (d, GB) is slightly inflamed.

alanine aminotransferase: 398.7 U/L, total bilirubin: 1.718 mg/dL, direct bilirubin: 1.463 mg/dL, gamma glutamyltranspeptidase: 241 U/L, alkaline phosphatase: 437 U/L. Echinococcus indirect fluorescent antibody testing and Echinococcus hemagglutination tests were positive. Contrast-enhanced axial computed tomography (CT) images through the liver showed that there was a cystic lesion in segment four communicating with the biliary system. The main hepatic duct was obliterated and dilated by curvilinear densities consistent with germinative membranes expelled from the cyst into the biliary system. Peripheral intrahepatic bile ducts were dilated as well. Gallbladder was slightly inflamed (Fig. 1d). The cyst was a thick-walled and partially collapsed one with no visible germinative membrane or any daughter cyst within and it was in communication with the bile ducts. On the other hand, the dilated bile ducts were completely obliterated by multilayered germinative membranes. The findings were consistent with a Gharbi Type II hydatid cyst, which had ruptured and expelled all the germinative membranes out into the bile ducts. Multiplanar reconstruction (MPR) CT image demonstrated the cystobiliary communication clearly and depicted the layering germinative membranes in the dilated extra hepatic bile ducts (Fig. 2). Chest X-ray revealed no cysts in the lung.

Patient's oral intake was stopped. Intravenous fluids, cefoperasone-sulbactam, and albendazole were

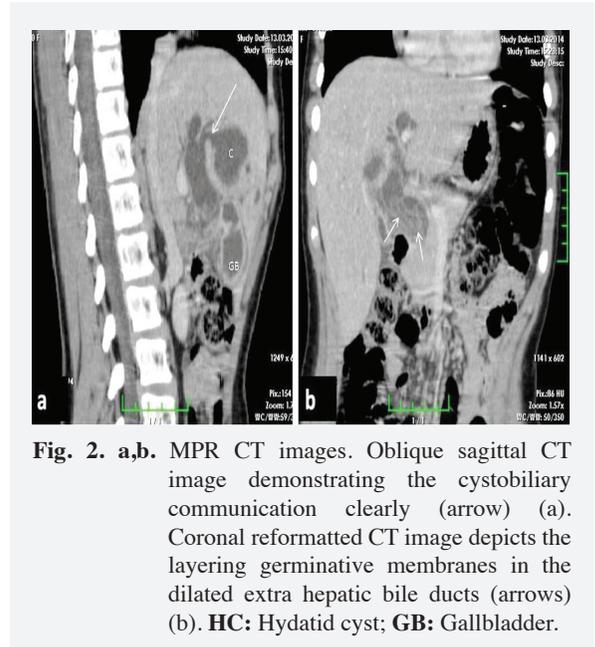


Fig. 2. a,b. MPR CT images. Oblique sagittal CT image demonstrating the cystobiliary communication clearly (arrow) (a). Coronal reformatted CT image depicts the layering germinative membranes in the dilated extra hepatic bile ducts (arrows) (b). **HC:** Hydatid cyst; **GB:** Gallbladder.

started. ERCP and sphincterotomy were planned because of the persistent choledochal obstruction findings after two days of conservative treatment. Patient was sedated by administration of midazolam and ketamine, and duodenal peristalsis was suppressed with hyoscine N-butyl bromide. The ERCP procedure was performed with patient in a prone position with video duodenoscopy (System 4400, Fujinon). Papilla was cannulated with a guide wire and standard papillotomy was performed. ERCP was performed with iohexole and showed multiple filling defects in the choledochus and a cyst pouch in the segment four of the liver (Fig. 3a). A 20-millimeter sphincterotomy was performed. Germinative membrane and daughter cysts were cleaned with the use of balloon and basket (Fig. 3b, c). No scolicalid agent was used during the procedure. Clearances of the cysts were confirmed with repeat iohexole imaging. Amoxicillin clavunate and albendazole were prescribed and patient discharged on postoperative first day without any complications. The patient admitted to our clinic with recurrent right upper abdomen pain on postoperative 4th day. Ultrasonography revealed mild obstruction in choledochus. Obstruction findings disappeared after two days of conservative treatment. Patient is doing well without any other complication 20 months after ERCP.

3. Discussion

Hydatid disease is a zoonosis which caused by larval forms of echinococcus tapeworms. Cystic echinococcosis is endemic mostly in sheep farming rural areas of the developing countries. Echinococcus granulosus is the most common form and gives rise to cysts primarily in the liver and lung. Echinococcus Multilocularis is much less common form and



Fig. 3. a-c. ERCP showing the cystic lesion communicating with the biliary tree and multiple filling defects in the extra hepatic bile ducts. Arrows show the germinative membranes (a). Germinative membrane that is removed through the duodenal papilla is seen (arrow) (b). ERCP showing the cystic lesion communicating with the biliary tree and filling defects in the extra hepatic bile ducts disappeared after cleaning balloon and basket (c). **ERCP:** Endoscopic retrograde cholangiopancreatography.

produces an invasive tumor-like replacement of liver tissue. Worldwide annual incidence of hydatid cyst is 1-200 per 100.000 inhabitants (Oral et al., 2012). However, the real worldwide incidence is unknown in children. In Turkey, the annual incidence in children is 150 cases per 100.000 children (Ok et al., 2007). Clinical symptoms may not occur until the cyst ruptured because of minor trauma. After the rupture if cyst leakage occurs, an allergic reaction may follow. Ultrasound examination is the most useful diagnostic test for hydatid cyst in abdominal locations (Oral et al., 2012). Chest roentgenogram is useful for lung involvement. In addition, CT and magnetic resonance imaging (MRI) are highly sensitive for diagnosis in the lung and abdomen. Although it has a low sensitivity, indirect hemagglutination (IHA) test is the most common serologic test in use (Oral et al., 2012).

Some complications can occur such as rupture, obstructive jaundice, acute cholangitis, thoracobiliary fistula, and intrabiliary fistula (Çetinkursun et al., 1996; Akaydin et al., 2012). Rupture of a hydatid cyst into the biliary tract, also known as cystobiliary communication, is the most common complication of a hepatic hydatid cyst. This may lead to obstructive jaundice, pancreatitis, cholangitis, and sepsis with high mortality (Wani et al., 2011). To determine the nature of the complication, ultrasonography, CT, MRI, and magnetic resonance cholangiopancreatography can be used as diagnostic methods. Surgery may be indicated for patients with symptoms and complications of hydatid disease (Akaydin et al., 2012). The specific

operative approach depends on the number and location of the cysts and associated complications. Endoscopic methods are reported to be effective alternatives to surgery for the treatment of such complications owing to lower morbidity and shorter hospital stay (Akaydin et al., 2012). In our case that had an acute presentation, ERCP helped us to both diagnose and treat the patient. Therapeutic approach consisted sphincterotomy and removal of germinative membrane and daughter cysts with the use of balloon and basket. No further treatment was required except albendazole to prevent recurrence. No scolicedal agent was used during the treatment in order to prevent damage to the biliary epithelium causing strictures. Sphincterotomy is being used to treat long-term biliary leakage after hydatid cyst surgery, but the use of ERCP with sphincterotomy alone for the treatment of ruptured hydatid cyst in to the biliary system is not a well-known procedure especially in countries where the disease is not endemic (Şahin et al., 2006).

Conclusion

There have been few case reports documenting the successful use of ERCP treatment of hydatid cyst in adult patients, and in our knowledge, it is the first paper describing ERCP treatment in a child in English literature (Dumas et al., 1999; Warman et al., 2002). ERCP may be a useful procedure in selected children with suspected intrabiliary rupture or common bile duct obstruction, which occurs due to daughter cyst and germinative membrane.

REFERENCES

Akaydin, M., Erozyen, F., Ersoy, Y.E., Birol, S., Kaplan, R., 2012. Treatment of hepatic hydatid disease complications using endoscopic retrograde cholangiopancreatography procedures. *Can. J. Surg.* 55, 244-248. doi: 10.1503/cjs.036010.

Akbulut, S., Senol, A., Sezgin, A., Cakabay, B., Dursun, M., Satici, O., 2010. Radical vs conservative surgery for hydatid liver

- cysts: Experience from single center. *World J. Gastroenterol.* 16, 953-959.
- Çetinkursun, S., Oztürk, H., Alper, A., Sakarya, M.T., Süre, İ., 1996. Kist hidatiğe bağlı torako bilier fistül. *Pediatric Cerrahi Dergisi.* 10,40-43.
- Dumas, R., Le Gall, P., Hastier, P., Buckley, M.J., Conio, M., Delmont, J.P., 1999. The role of endoscopic retrograde cholangiopancreatography in the management of hepatic hydatid disease. *Endoscopy.* 31, 242-247.
- Mandelia, A., Wahal, A., Solanki, S., Srinivas, M., Bhatnagar, V., 2012. Pancreatic hydatid cyst masquerading as a choledochal cyst. *J. Pediatr. Surg.* 47, 41-44. doi: 10.1016/j.jpedsurg.2012.07.054.
- Ok, U.Z., Ozkol, M., Kilimcioğlu, A.A., Dinç, G., Bayindir, P., Ostan, I., Pabuşçu, Y., Ozcan, C., Korkmaz, M., Coşkun, S., Yüksel, H., Girginkardeşler, N., 2007. A province-based study using sampling method to investigate the prevalence of cystic echinococcosis among primary school children in Manisa, Turkey. *Acta Trop.* 103, 116-122.
- Oral, A., Yigiter, M., Yildiz, A., Yalcin, O., Dikmen, T., Eren, S., Kantarci, M., Salman, A.B., 2012. Diagnosis and management of hydatid liver disease in children: A report of 156 patients with hydatid disease. *J. Pediatr. Surg.* 47, 528-534. doi: 10.1016/j.jpedsurg.2011.11.007.
- Şahin, D. A., Kuşaslan, R., Türel, K.S., Akbulut, G., Arıkan, Y., Dilek, O. N., 2006. Surgical treatment in our hydatid cyst patients and efficiency of sphincterotomy with ERCP. *The Med. J. Kocatepe.* 6, 11-16.
- Wani, N. A., Kosar, T., Gojwari, T., Robbani, I., Choh, N.A., Shah, A.I., Khan, A.Q., 2011. Intrabiliary rupture of hepatic hydatid cyst: Multidetector-row CT demonstration. *Abdom Imaging.* 36, 433-437. doi: 10.1007/s00261-010-9675-8.
- Warman, S., Meier, P.N., Kardorff, R., Fuchs, J., 2002. Cystic echinococcosis with perforation into the biliary tract in an eight-year-old girl. *Eur. J. Pediatr. Surg.* 12, 134-137.