

## HYDATID DISEASE IN CHILDREN AND BILIARY FISTULAS

**Zoran Marjanovic**

Faculty of Medicine, University in Nis, Serbia [drzmarjanovic@gmail.com](mailto:drzmarjanovic@gmail.com)

**Andjelka Slavkovic**

Faculty of Medicine, University in Nis, Serbia

**Dragoljub Zivanovic**

Faculty of Medicine, University in Nis, Serbia

**Ivona Djordjevic**

Faculty of Medicine, University in Nis, Serbia

**Abstract:** Echinococcosis is a parasitic infection caused by the larva of *Echinococcus granulosus* and is endemic in many countries where it also represents a major public health problem. There are many different surgical techniques for the management of echinococcosis described, and most common and serious complication of liver cyst management is occurrence of biliary fistula. We present two cases with biliary fistulas that are managed conservatively in a non-endemic region center. Treatment of biliary fistulas in children is underreported unlike in the adults where different modalities of prevention and fistula management have been widely described. Although there are various techniques reported, treatment of hydatid disease and its complications is complex and challenging especially in pediatric age group.

**Keywords:** children, hydatid disease, biliary fistula.

### 1. INTRODUCTION

Human hydatid disease or echinococcosis is a parasitic infection caused by the larva of *Echinococcus granulosus* and has worldwide distribution and is endemic in many countries [1-4]. The liver is the most common site of involvement (70-75%), followed by the lungs (15%); localization on other organs is rare [1,3,5]. Although Albendazole is used in medical treatment of Echinococcosis, only 31.5% patients were considered as cured [6]. Surgery remains a cornerstone of hydatid cyst treatment with cystobiliar communication as the most common complication. Biliary fistula develops when the postoperative leak is able to drain; if it cannot, biliary peritonitis and biliary abscess develop [7].

### 2. METHODS

This article presents three cases of liver hydatid cysts in children who underwent cystectomy and pericystectomy where a high-output (>300ml/24h) biliary fistula developed. Emphasis is placed on the importance of the decision of whether to wait for spontaneous closing of fistula or to treat it surgically in a non-endemic center with small number of patients.

### 3. RESULTS

A total of 24 patients underwent surgery for hydatid cysts in a 10-year period (2005 – 2015) in our center. A retrospective study was performed to evaluate surgical management and complications in the non-endemic region.

In the 10-year period, there were 24 children treated for echinococcosis in our center; 15 boys and 9 girls with mean age of 8.83 years (2-17) at surgery. In 20 patients liver was affected. Cystectomy with partial pericystectomy was performed in n=22, n=1 underwent puncture-aspiration-injection-reaspiration (PAIR) and n=1 partial liver resection. Biliary fistula occurred as a complication in three patients.

Case one: First patient was a 13-year-old girl. *Echinococcus* solitary liver cyst was first diagnosed by ultrasound examination and cyst measuring 10 cm was discovered in VI and VII liver segment. MRI findings confirmed initial US diagnosis (Figure 1). Patient underwent cystectomy with partial pericystectomy without any intraoperative complications. On the second postoperative day biliary fistula occurred. Biliary drainage was from 90 to 370ml per day, with average drainage 175ml/24h (Figure 2). After 35 days from surgery there was no bile leakage and abdominal drain was removed four days after drainage stopped. There was no recurrence and control MRI showed no cyst of biliary leakage.

Thirteenth International Scientific Conference  
THE TEACHER OF THE FUTURE  
25-28.5.2017, Budva, Montenegro

Case two: Second patient was 9 years old boy with solitary liver cyst in right liver lobe. Ultrasound and MRI were performed and cyst with dimensions 55x39x52 mm in VI and partially V liver segment was described. Biliary fistula occurred two days after surgery. Biliary drainage was from 70 to 450 ml per day with average drainage of 250ml/24h. Biliary fistula persisted for 31 days (Figure 3), and three days after no bile drainage was registered, the abdominal drain was removed. Control ultrasonographic and MRI findings showed no recurrence.

Case three: A 10-year-old boy was admitted for further diagnosis and treatment, due to US finding of ascites and splenomegaly, and laboratory findings of initial liver dysfunction (AST 116, ALT 53, ALP 254, proteins 57, albumins 29). Repeated US and MSCT confirmed cyst within porta hepatis (47x40 mm), just between left and right common bile duct, but showed also dilation of intrahepatic biliary ducts (Figure 3). Patient underwent cystectomy with partial pericystectomy, followed by the drainage of cystic cavity. On the third day, a biliary fistula occurred, initially low-, but later highproductive, with maximum of 600 ml/24h (average 325ml/24h). After 40th day, the secretion reduced in volume, and on the 56th day, the drainage finally stopped (Figure 4).

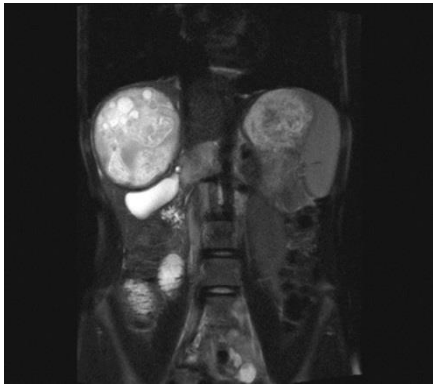


Figure 1. MRI showing liver cyst

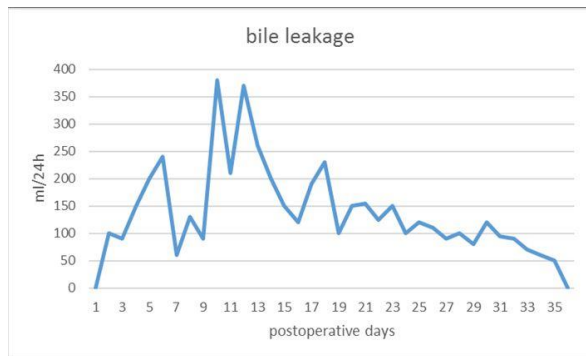


Figure 2. Postoperative bile leakage

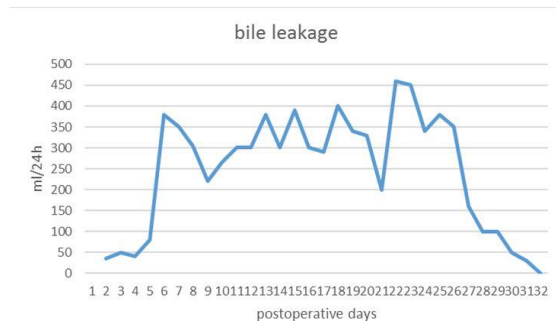


Figure 3. Postoperative bile leakage

#### 4. DISCUSSION

Thirteenth International Scientific Conference  
THE TEACHER OF THE FUTURE  
25-28.5.2017, Budva, Montenegro

---

Echinococcosis is caused by the larval form of *Echinococcus granulosus* and is usually presented as uncomplicated cyst (82%), liver is most affected organ; right lobe in 80.6%, left lobe in 6.5% and bilaterally in 12.9% [1,2]. Ultrasound is the most common diagnostic method, CT scan and serology are useful for differential diagnosis. Surgical treatment can be radical or conservative. Radical procedures are pericystectomy or hepatic resection. Conservative techniques are deroofting of cyst and management of the residual cavity by external drainage, omentopexy, capsulorrhaphy, capitonnage and marsupialization [4]. Also, laparoscopic approaches and PAIR are used for surgical treatment. Scolicidal solutions may be used in surgical (and percutaneous) approaches [1]. The most frequent complication of hepatic hydatid cyst treatment is biliary fistula. Common symptoms in patients with external biliary fistula are jaundice, cholangitis or biliary colic [3]. Cystobiliary communication can be frank (5-17%) or occult. Frank communications are easily diagnosed preoperatively, occult fistulas arise as external biliary fistula, biliary peritonitis or biliary abscess in postoperative period. Although clear cystic fluid is believed to indicate noncommunication with the biliary system, Sönmez et al. reported two cases where bile leak occurred after aspiration of clear cystic fluid [1,7]. Histopathological analysis confirmed that patients with large fistulas had chronic sclerosing cholangitis and dilatation of smaller bile ducts. Dilatation of small bile ducts is rightly considered as precursor sign for large fistulas [8]. There are several surgical options and surgeons often prefer the technique they know the best; placement of surgical drains remains a surgeon's choice. As abdominal drains were placed after cystectomies and pericystectomies in our patients, it helped us to follow up bile leakage per day as decision was to wait first for fistulas to close spontaneously. Decision was made as endoscopic retrograde cholangiopancreatography (ERCP) is not routinely done in our hospital. Farhat et al. reported ERCP in management of biliary fistulas in 26 adult patients where the mean time from initial ERCP to running dry of the leaks was  $17.5 \pm 12.4$  days and no complications have been reported [9]. The role of ERCP in the pediatric age group is not well established, because pancreatic and biliary diseases are less common in children. Issa et al reported ERCP as a diagnostic and therapeutic procedure in 125 children with biliary and pancreatic disorders, three of them had bile leak and this is one of the largest series in pediatric age group [10]. Different fistula management and prevention are described. Roux-en-Y cystojejunostomy as a management method for external biliary fistula is reported by Mosaddeghi et al. [4]. Özmen and Coşkun have suggested the use of a telescope during surgery to determine the relationship between the bile ducts and the cyst [7]. Mosaddeghi et al reported capsulorrhaphy technique in 250 patients, biliary fistula occurred in only one patient (0,4%) which is the lowest reported rate [4].

## 5. CONCLUSION

We can conclude that there is no unique approach for prevention and management of biliary fistulas in children with liver hydatid disease. If patients are treated in non endemic countries with small number of cases, capsulorrhaphy is a good choice to avoid biliary complications. On the other hand, ERCP is reported as a safe and efficient method for treatment in those centers where it is routinely done.

## REFERENCES

- [1] Sönmez K, Karabulut R, Türkyılmaz Z, Can Basaklar A, Kale N. Clear Cystic Fluid in Hepatic Hydatidosis Does Not Rule Out Communication Between Cysts and the Biliary System. *Adv Ther* 2007;24(2):291-5
- [2] Celebi F, Balik AA, Salman AB, Oren D. Hydatid disease in childhood. *Pediatr Surg Int* 2002;18(5-6):417-9
- [3] Akaydin M, Erozen F, Ersoy YE, Birol S, Kaplan R. Treatment of hepatic hydatid disease complications using endoscopic retrograde cholangiopancreatography procedures. *Can J Surg* 2012;55(4):244-8
- [4] Mosaddeghi KS, Heris HK, Bayat A, Mosaddeghi KZ. Capsulorrhaphy in the management of liver hydatid cyst. *Ann Hepatol* 2014;13(3):378-83
- [5] Sharma BC, Reddy RS, Garg V. Endoscopic management of hepatic hydatid cyst with biliary communication. *Dig Endosc* 2012;24(4):367-70
- [6] Ben Brahim M, Nouri A, Ksia A, El Ezzi O, Krichene I, Mekki M, et al. Management of multiple echinococcosis in childhood with albendazole and surgery. *J Pediatr Surg* 2008;43(11):2024-30
- [7] Atahan K, Küpeli H, Deniz M, Gür S, Çökmez A, Tarcan E. Can Occult Cystobiliary Fistulas in Hepatic Hydatid Disease Be Predicted Before Surgery? *Int J Med Sci* 2011;8(4):315-20
- [8] Stamm B, Fejgl M, Hueber C. Satellite cysts and biliary fistulas in hydatid liver disease. A retrospective study of 17 liver resections. *Hum Pathol* 2008;39(2):231-5

Thirteenth International Scientific Conference  
THE TEACHER OF THE FUTURE  
25-28.5.2017, Budva, Montenegro

---

- [9] Farhat S, Bourrier A, Gaudric M, Dousset B, Scatton O, Chaussade S, Prat F. Endoscopic treatment of biliary fistulas after complex liver resection. *Ann Surg.* 2011;253(1):88-93
- [10] Issa H, Al-Haddad A, Al-Salem AH. Diagnostic and therapeutic ERCP in the pediatric age group. *Pediatr Surg Int* 2007;23(2):111-6