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ERCP treatment of obstructive jaundice caused by hydatid cyst in extrahepatic ducts 13 years after liver hydatid endocystectomy. A case report

Astrit Hamza [a,b,d], Avdyl Krasniqi [d], Fatos Sada [b,c,d], Valon Zejnullahu [a], Besnik Bicaj [b,d,*]

[a] Department of Abdominal Surgery, Clinic of Surgery, University Clinical Centre of Kosova (UCCK), 10000, Prishtina, Kosovo
[b] Faculty of Medicine, University of Prishtina, 10000, Prishtina, Kosovo
[c] Department of Anesthesiology and Reanimation, University Clinical Centre of Kosova (UCCK), 10000, Prishtina, Kosovo
[d] Kavaja Hospital, Prishtina-Ferizaj highway 5th km, 10000, Prishtina, Kosovo

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ABSTRACT

INTRODUCTION: Hydatid disease affects most commonly the liver, and complications with the rupture into the biliary tree develop in approximately one-fourth of the cases. Moreover, primary hydatid cysts of the biliary tract have been reported.
PRESENTATION OF CASE: We report an extremely rare case of obstructive jaundice caused by hydatid cyst in extrahepatic ducts 13 years after liver hydatid endocystectomy treated by Endoscopic retrograde cholangiopancreatography (ERCP). A 28-year-old male patient who had undergone surgical treatment – removal of liver hydatid cyst 13 years earlier, presented with signs of obstructed jaundice, confirmed with blood tests results and magnetic resonance cholangiopancreatography (MRCP). Actually, there were no pathological changes detected in the hepatic parenchyma, but the intrahepatic and extrahepatic bile ducts were dilated. ERCP was performed and the entire hydatid material was evacuated and washed out into the gastrointestinal tract. In addition, after laparoscopic cholecystectomy, hydatid cysts were also confirmed in the gallbladder.
DISCUSSION: Generally, the obstructive jaundice caused by hydatid cyst in the extrahepatic ducts can also be caused by the rupture of the liver hydatid cyst in the biliary tract, or by primary hydatid cyst in the biliary tract. The ERCP plays a key role in the diagnosis and the treatment of this pathology.
CONCLUSION: The ERCP, has now become an important diagnostic and therapeutic procedure in management of primary extrahepatic hydatid cysts and of complicated liver hydatid cysts.

1. Introduction

Hydatid disease is a major endemic health problem in certain areas of the world [1]. Hydatid disease is a parasitic infestation by a tapeworm of the genus *Echinococcus* which belongs to the Taeniidae family. There are four species of *Echinococcus*: *E* granulosus, E. multilocularis, E. vogeli and E. oligarthrus. The first two species are the most common, causing cystic echinococcosis and alveolar echinococcosis, while the third and fourth cause polycystic echinococcosis, but have only rarely been associated with human infection [2–4].

Humans represent intermediate host that are infested through ingestion of parasite eggs found in contaminated food, water, or soil, or after direct contact with animal hosts. After humans' being infested, 50%–75% of hydatid cysts occur in the liver, 25 % are found in lungs, and 5%–10% are distributed to other organs through the arterial system [5]. However, in approximately one-fourth of the cases, hepatic hydatid cyst ruptures into the biliary tree (because of higher pressure in the cyst, often up to 80 cm H2O), producing obstructive jaundice [6].

There are two types of cystic rupture into the biliary tree: frank intrabiliary rupture, and simple communication. The patients with frank intrabiliary ruptures are usually symptomatic and treatment methods are tailored at the same time either with surgery or with endoscopic procedures. By contrast, the patients with simple communications may be asymptomatic, with the communications identified only during surgery, or even remain undetected.

Moreover, primary hydatid cysts of the biliary tract have been reported [7]. We are presenting an extremely rare case of obstructive jaundice caused by hydatid cysts in extrahepatic ducts 13 years

Abbreviations: ERCP, endoscopic retrograde cholangiopancreatography; MRCP, magnetic resonance cholangiopancreatography; CT, computed tomography; CBD, common bile duct.

* Corresponding author at: Faculty of Medicine, University of Prishtina, Kavaja Hospital, Prishtina-Ferizaj highway 5th km, 10000, Prishtina, Kosovo.
E-mail address: besnik.bicaj@uni-pr.edu (B. Bicaj).

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after liver hydatid endocystectomy treated by the ERCP. This case is reported in line with the SCARE criteria [8].

2. Presentation of case

A 28-year-old Caucasian male patient admitted in our hospital with complaints of constant mild pain in the right upper abdomen, accompanied by nausea and jaundice. There was no history of high degree fever. In previous medical history, he had an uncomplicated liver hydatid endocystectomy 13 years earlier. Physical examination showed mild tenderness in the right upper quadrant of the abdomen where the right subcostal incision was noticed. Laboratory findings revealed increase in serum aspartate transaminase (106 U/L) and alanine aminotransferase levels (109 U/L), elevated bilirubin (total bilirubin/direct bilirubin 61/22 μ mol/L), Alkaline phosphatase (823 U/L), and gamma glutamyl transferase (270 U/L). All other laboratory examinations were normal. The ultrasound of the abdomen showed a mass in the extrahepatic duct with dilation of the intrahepatic biliary ducts. Hepatic parenchyma was without pathological changes. Gallbladder was hypoechogenic with a well-defined intraluminal mass, kind of biliary sludge without stone or wall thickening. The MRI and MRCP revealed extensively dilated hepatic bile ducts and common bile duct filled with hydatid cyst material seen as hypointense lesions (Fig. 1).

In order to confirm the diagnosis and to remove the material from the biliary tree, an ERCP was indicated. An endoscopic sphincterotomy was performed and a large amount of the membranes and daughter vesicles were removed from the common bile duct (CBD) extraction by balloon and basket catheter (Fig. 2). After emptying hydatid membranes and big daughter vesicles, CBD was irrigated with saline flushing out the hydatid material and small daughter cysts. At the same time the patient was sent to the operating room and a laparoscopic cholecystectomy was performed. After cholecystectomy, the gallbladder was opened confirming also membranes and small daughter cysts inside gallbladder (Fig. 3). Perioperative course of the patient was uneventful. Due to multiple ERCP applications with balloon and basket catheter in CBD and in order to prevent any cholangitis, the patient was treated with antibiotics (Ceftriaxone 1 g IV twice daily) for 7 days. On the second postoperative day, he was discharged from hospital in good condition. However, due to suspicions regarding systemic infections, Albendazole 400 mg BID was also prescribed for eight weeks. The patient recovered subsequently with decrease in jaundice, fall of total serum bilirubin to normal range. The patient remained asymptomatic during the six months of follow up.

3. Discussion

It remains an undisputable fact that high percentage of hydatid cysts in the liver, besides other organs, are caused by infestation by a tapeworm of the genus Echinococcus [5]. Over the past decades, surgical management was one of the best treatment method for liver hydatid disease [9]. However, with the advancements in chemotherapy and percutaneous therapy techniques, surgery is reserved mainly for the complicated hydatid cysts [4,10]. Despite advances in surgical techniques and the use of chemotherapy, recurrence remains one of the major problems in the management of liver hydatid disease ranging from 4.6%–22.0 % [11,12]. However, most of the recurrence cases reported in literature in English occurred within two years after operation [13]. On the other hand, based on treatment modality, the recurrence may occur decades after surgery [11,14]. According to our patient's history and the medical records, he had uncomplicated liver hydatid endocystectomy 13 years earlier. At that time, there was either no communication between the hydatid cyst and the biliary tree, or, if it was present, it was not detected. After the surgery, the patient was treated with Albendazole 400 mg BID for eight weeks. However, in approximately one-fourth of the cases, hepatic hydatid cyst ruptures into the biliary tree (due to higher pressure in the cyst, often up to 80 cm H2O), producing obstructive jaundice [6].

We could speculate that obstructive jaundice caused by hydatid cyst in extrahepatic ducts in our case may have occurred due to any previous simple cysto-biliary communication. It has appeared clinically with jaundice 13 years later; however, the likelihood of this to happen is minimal. To our best knowledge, and based on the literature published in English, an obstructive jaundice caused by hydatid cyst in extrahepatic ducts after liver hydatid endocystectomy after a long period of time, similar to our case, cannot be found. On the other hand, primary hydatid cysts of the biliary tract have been reported earlier [7]; such a case was also reported by our team [15]. The pathogenesis of the unusual locations of hydatid cysts support the hypothesis that beside portal circulation, the echinococcus embryos can spread via other routes, such as the lymphatic system,

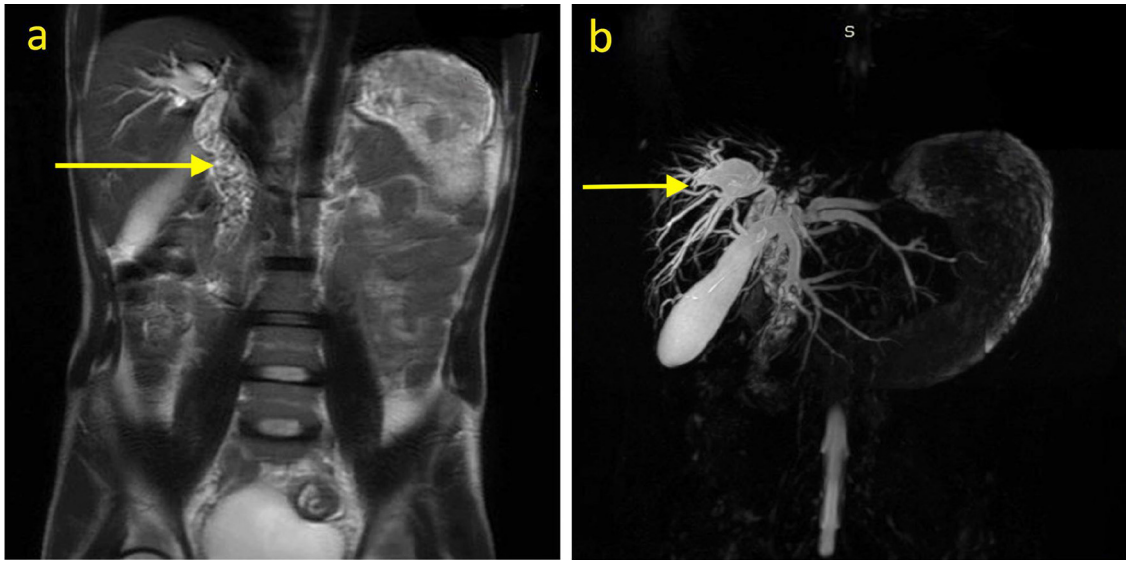

Fig. 1. MRI (a) demonstrating extensively dilated common bile duct filled with hydatid cyst material seen as hypointense lesions (indicated by the yellow arrow). and MRCP (b) demonstrating dilated intrahepatic bile ducts (indicated by the yellow arrow).

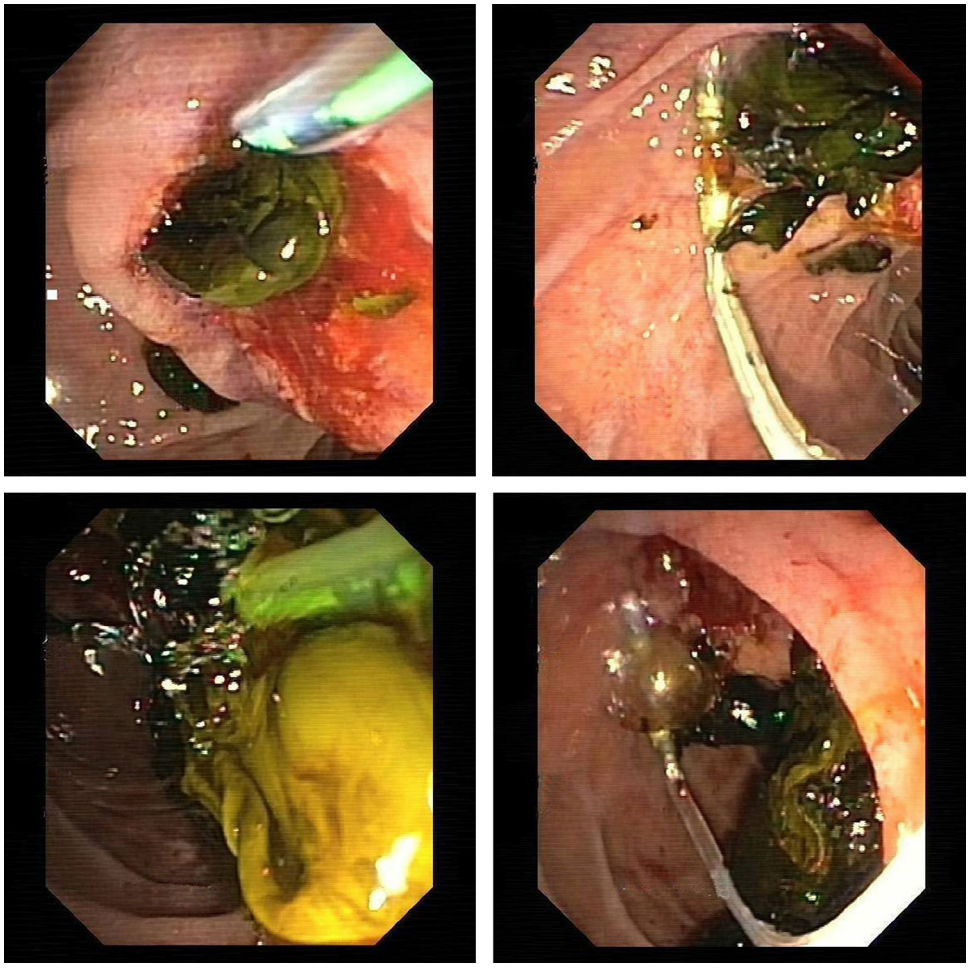

Fig. 2. Endoscopic retrograde cholangiopancreatography showing evacuation of hydatid membranes and daughter cysts with endoscopic sphincterotomy and a biliary occlusion balloon.

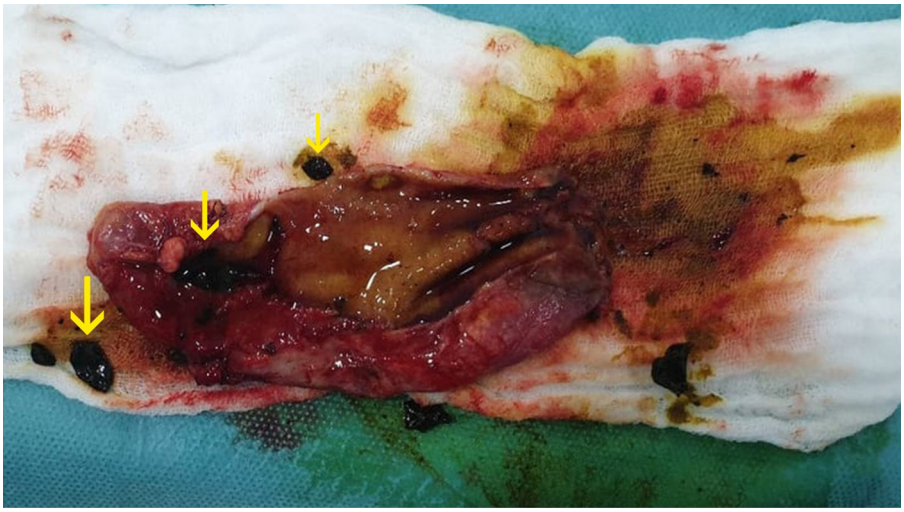

Fig. 3. Open gallbladder after cholecystectomy where hydatid membrane and daughter cysts where noticed (arrows).

which further extend to involve the luminal mucosa and the biliary tract, although the embryos could invade the biliary lumen directly [15–17]. In our case, hydatid cyst, in addition to being in the bile ducts, was also present in the gallbladder. Hence, the location of the hydatid cyst in the biliary tree and the gallbladder could happen by the same mechanisms elaborated above. ERCP has become the primary diagnostic and therapeutic method of primary hydatid cyst in the biliary tree, which has also been successful in addressing other complications of biliary hydatidosis such as clearing the biliary tree, closing fistulas and biliary leaks [3,10,18,19].

A sphincterotomy is often needed in patients with obstructive jaundice and the cysts, and membranes can then be removed with the help of a basket or an occlusion balloon [10]. Saline irrigation of the bile duct may be necessary to flush out the hydatid sand and

small daughter cysts. We performed the same procedure followed by laparoscopic cholecystectomy where the hydatid material and daughter cysts were confirmed in the gallbladder.

4. Conclusion

ERCP represents an important management strategy for patients with hydatid cysts in extra biliary tree, either primary or complications of liver hydatid cyst, such as clearing the biliary tree, closing fistulas and biliary leaks, followed by laparoscopic cholecystectomy, when the hydatid cyst is also located in the gallbladder. Moreover, based on our team experience in the treatment of this disease, preoperative MRCP is required to diagnose complicated liver hydatid cysts in order that the treatment modality be done in the right time.

Declaration of Competing Interest

The authors have no conflicts of interests.

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This case report is realized without any founding.

Ethical approval

There was no ethics approval required for this case report.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

AH, AK, BB and FS participated in the Endoscopy, Laparoscopy and anaesthesiology of this case. AH, BB and VZ treated the patient after surgery. AH drafted the manuscript and all authors read and approved the final manuscript.

Registration of research studies

Not applicable.

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