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Biliary Obstruction as a Late Complication after PAIR Treatment of Liver Hydatidosis: Case Report

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Abstract

Background: Hydatid liver disease could complicate the perforation into biliary ducts with the formation of cystobiliary communication (CBC) or this may occur like a complication of the percutaneous or surgical treatment. CBC complicated with biliary obstruction is extremely rare condition. There are only 15 reports about obstructive cholangiohydatidosis and secondary cholangitis in English literature. To the best of our knowledge, our report is the first case report of obstructive cholangiohydatidosis as a late complication of the PAIR treatment of liver hydatidosis.

Case summary: The authors present a successful treated case of liver hydatid cyst type CE3A, according to the WHO classification, presented with obstructive jaundice which occurred one month after PAIR procedure. Open surgical choledochotomy was performed, and scolex cysts and debris into the common bile duct were found and evacuated. Catheter external-internal biliary drainage and additional partial peri cystectomy of the hydatide cyst was performed. The patient recovered well from the operation and his postoperative course was uneventful.

Conclusions: This case report suggest that intra-biliary rupture and scolex obstruction might be a complicated natural course of necrotic changes in hydatide cyst and biliary tree and missed during PAIR procedure, either occurred iatrogenic ally during the implementation of a therapeutic procedure. CBC requires an early diagnosis, and a decision on further treatment, for a better prognosis of the disease. Surgery, however, is the gold standard in the treatment of hydatid liver disease, as well as in the treatment of its complications.

Keywords: Biliary Obstruction; Cholangitis; Complication; PAIR; Liver; Hydatidosis

Abbreviations: CBC: Cystobiliary Communication; LH: Liver Hydatidosi; MRI: Magnetic Resonance Imaging; MRCP: Magnetic Resonance Cholangiopancreatography; INCH: Intraoperative Cholangiography

Introduction

Hydatid disease or Echinococcosis is an endemic zoonosis of the Mediterranean region, Africa, South America, Middle East, Australia and New Zealand. It is in most cases located in the liver [1-3]. Liver hydatidosi (LH) could complicate with the perforation into biliary ducts with the formation of cystobiliary communication (CBC), rupture into the peritoneal cavity, and bronchi [4], cyst infection and other rare complications. Treatment of hydatid cyst of the liver can vary from surgical intervention to percutaneous drainage to medical therapy. At present, percutaneous intervention like PAIR (Puncture of the cyst, Aspiration of the cystic fluid, Injection of hypertonic saline, and Respiration of solution) is generally indicated for type CE1 (pure fluid collection), type CE3A (cyst with detached membranes), and some type CE3B (fluid collection with multiseptated cysts with daughter cysts) [5]. In most other cases, open or laparoscopic surgery is still considered the gold standard treatment. We present a successful treated case of liver hydatid cyst type CE3A, according to the WHO classification, presented with obstructive jaundice which occurred one month after PAIR procedure. Biliary obstruction was probably caused by echinococcus scolex and debris migrated to major bile ducts trough silent unrecognized primary cystobiliary communication (CBC), or secondary CBC created iatrogenic during the performance of the PAIR procedure.

Case presentation

Chief Complaints

We report the case of a 70-year-old man referred to our institution -tertiary Hepato-biliary University Clinical Center with signs of obstructive jaundice, one month after percutaneous method of treating liver hydatidosis (PAIR procedure) performed in regional institution.

History of Past Illness

The complete medical history from previous hospitalization was sent to us from regional hospital showed: Routine ultrasound of the abdomen in asymptomatic patient performed 6 months ago, revealed two cystic lesions in the right liver. Magnetic Resonance Imaging (MRI) of the abdomen verified two clearly limited cystic changes in segment 5 and segment 6 measuring 40x30mm and 57x46mm communicated with each other. Within the cysts, the liquid matrix shows irregular spiral band formations that are primarily due to the detached germinal membrane (CE3A) . Intra and extrahepatic bile ducts were not dilated, and no pathological content was visible in the lumen (Figure 1A). Serological test to Echinococcus granulosus was negative. Despite of that fact, the case was diagnosed like liver hydatidosis type CE3A according to WHO classification. It was treated using Albendazole 400mg a day for 28 days, 14 days off (three cycles), with no complication. After 5 months, the patient was referred to an interventional radiologist to evaluate the possibility of percutaneous treatment. PAIR procedure was performed.

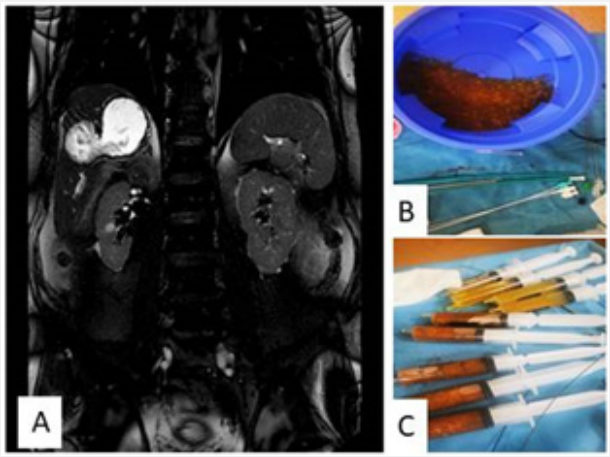

Figure 1: MRI of the abdomen [A]; Cystic content [B]; The initial aspirate forwarded for cytological and biochemical and microbiological analyses [C].

A cystic change in the right lobe of the liver was punctured under ultrasound control and a 10F diameter drainage catheter was placed intraluminally. The aspirated content was thick and gelatinous (Figure 1B). Then, by rotating the catheter itself inside the cyst, partial peeling of the walls and membranes and aspiration was performed. The initial aspirate forwarded for cytological and biochemical and microbiological analyses (Figure 1C), but operator did not take time to wait for the results. During this procedure contrast X-ray imaging was not performed. Sclerosation was performed with appropriate volume of absolute alcohol. Immediate post-interventional course was uneventful. On the 7th day after the PAIR procedure, the previously placed drainage catheter was extracted. A month later, jaundice, subfebrile temperature and generalized body weakness appeared, and patient was urgently referred to our institution.

Physical Examination

On physical examination, yellow discoloration of the skin and visible mucous membranes, and subfebrile body temperature (37,7 ^{0}C) were registered, with no other remarkable signs.

Laboratory Testing

Biochemical and hematological tests revealed severe transaminase Mia and hyperbilirubinemia (AST 224 U/L; ALT 196U/L; bilirubin 230.8 umol/L, direct bilirubin 121.8 umol/L; alkaline phosphatase - 444 U/L; gamma-glutamyl transferase (γ-GT) 608 U/L; LDH 608 U/L). Biochemical parameters of inflammatory syndrome occurred with elevated WBC (18 G/L) and CRP (130 U/L).

Imaging Examination

Magnetic resonance cholangiopancreatography (MRCP) revealed obvious communication between hydatide cyst and biliary tree at the level of anterior sectional bile duct (Figure 2).

Multidisciplinary Expert Consultation and Final Diagnosis

The multidisciplinary team made a diagnosis of biliary obstruction caused by hyda-tide debris and scolex passed from hydatide cyst, trough cystobiliary communication into the biliary

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tree. Because of high degree of the icterus and signs of cholangitis, urgent intervention was indicated. Surgery was chosen instead of endoscopy because of severity of symptoms and due to suspicion of incomplete cyst evacuation during the PAIR procedure.

Treatment

Patient underwent right subcostal laparotomy. After cholecystectomy, intraoperative ultrasound was performed, with clear vision of the hydatide cyst, dilated bile ducts and suspected communication. between them (Figure 3A). Intraoperative cholangiography [INCH] was performed through the cystic duct, which showed extravasation of the contrast agent from the right anterior sectional bile duct into the cystic cavity trough communication between these two structures. Distally, there was not any obstruction in the bile duct. The contrast passed to duodenum with no resistance (Figure 3B).

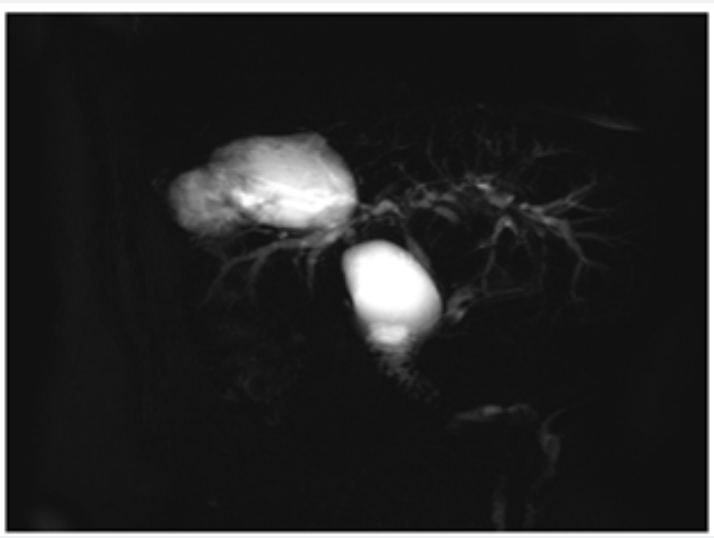

Figure 2: MRCP of cysto-biliary communication between hydatide cyst and biliary tree at the level of anterior sectional bile duct.

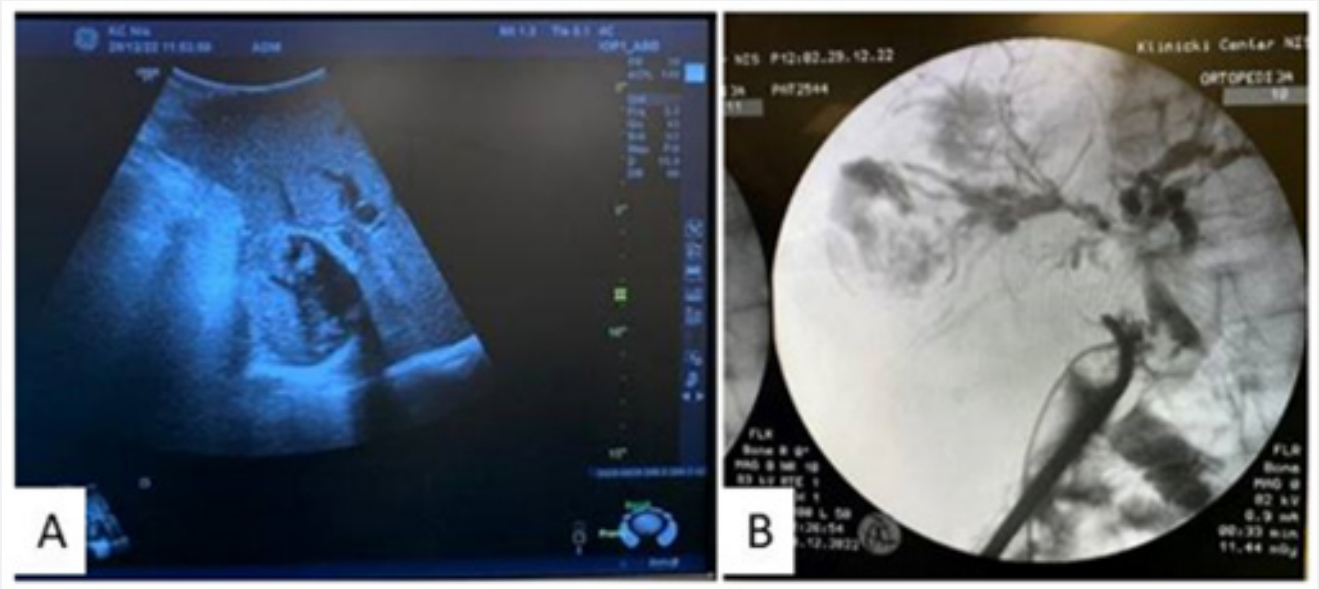

Figure 3: Intraoperative ultrasound (A) and trans cystic cholangiography (B).

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Choledochotomy was performed, and scolex cysts and debris into the common bile duct were found and evacuated (Figure 4A & 4B). The biliary catheter was inserted into the right bile duct, where the tip of the biliary catheter appeared extra hepatic at the level of the echinococcal cyst (Figure 4C). The proximal part of the catheter was externalized to the anterior abdominal wall as external biliary drainage, and the distal part remained in the common bile duct (external-internal drainage). Additionally, partial peri cystectomy of the hydatide cyst was performed. The right subphrenic and subhepatic space were drained.

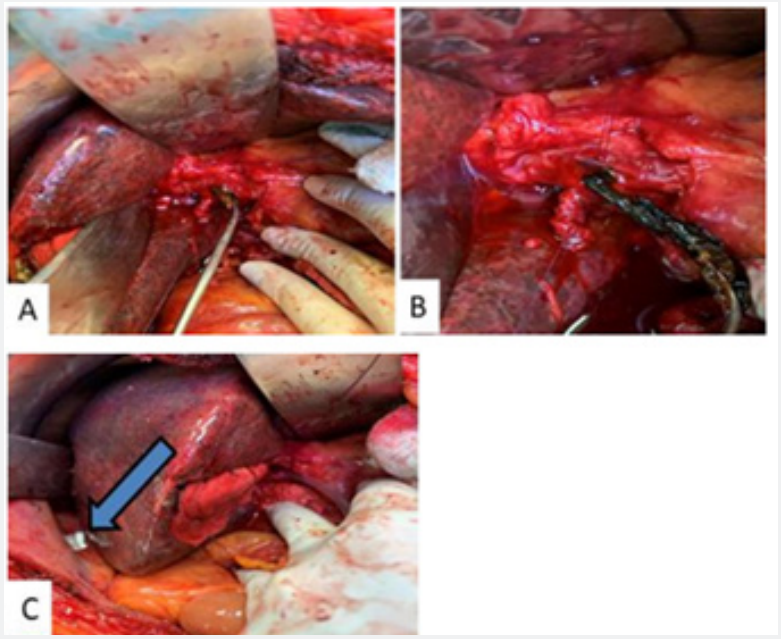

Figure 4: Intraoperative images- Opened common bile duct (A). Scolex cysts and debris (B). Fistulous communication with the right bile duct in which biliary catheter (arrow) has been passed and externalized (C).

Outcome and Follow-Up

The patient recovered well from the operation and his postoperative course was un-eventful. He was discharged from the hospital 14 days after the operation. The biliary catheter was removed after a control cholangiography was done on day 13th postoperatively.

Discussion

Hydatid disease is a worldwide zoonosis produced by the larval stage of the Echinococcus tapeworm. The two main types of hydatid disease are caused mostly by *E. granulosus* and less often by *E. multilocular* is [6]. According to WHO, the incidence of hyda-tide disease ranges from 1 to 200 cases per 100.000 populations[7]. *E. granulosus* is endemic disease in the great grazing regions like Mediterranean region, Africa, South America, Middle East, Australia, New Zealand. Man serves as intermediate host, being infected by ingestion of food contaminated by eggs excreted by the definitive host [canine]. The liver acts as the first filter in the disease and lungs act as the second filter. Because of that over 75% of hydatid disease are situated in the liver -liver hydatidosis (LH). Hydatid cysts may remain asymptomatic for many years and may be found incidentally on imaging [3,8]. In most cases, symptoms is coming like results of LH complication. Complications are registered in 30% and the most important are inflammation (abscess), hemorrhage, and rupture into the bile ducts, abdominal cavity or chest cavity.

However, some complications can even lead to fatal outcomes, for example, anaphylaxis after a cyst's rupture into the peritoneum or into the biliary system [8]. One of the most frequent and serious complications of LH is silent rupture into the bile ducts, with formation of CBC. This communication is most frequently discovered during surgery. In small percent of the patients, scolex and daughter cysts could migrate through the CBC into the biliary tree. This, so called "cholangiohydatidosis" can cause: biliary obstructive syndrome -mechanical icterus, acute suppurated cholangitis, and even acute secondary pancreatitis[9]. The diagnosis of LH is made on the basis of history, clinical examination, serological tests and imaging procedures. On the basis of ultrasound, liver hydatidosis can be classified according to Gharbi and WHO -IWGE classification (Table 1) [10-12]. MR and CT are methods used for more precise localization and delineation of the anatomy, necessary for planning surgical or radiological intervention. MRCP is mandatory in the cases of suspected complication, in particular biliary invasion and CBC.

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Table 1: Comparative description of the WHO-IWGE and Gharbi ultrasound classifications of echinococcal cysts.

WHO-IWGE 2001	Gharbi 1981	Description	Stage	PAIR [indication]
CE1	Type I	Unilocular unechoic cystic lesion with double line sign.	Active	Indicated
CE2	Type III	Multiseptated "rosette-like" "honeycomb" cyst.	Active	Contraindicated
CE3 A	Type II	Cyst with detached membranes [water-lily-sign].	Transitional	Indicated
CE3 B	Type III	Cyst with daughter cysts in solid matrix.	Transitional	Contraindicated
CE4	Type IV	Cyst with heterogenous contents. No daughter cysts.	Inactive	Contraindicated
CE5	Type V	Solid cyst with calcified wall	Inactive	Contraindicated

There are various treatment modalities for this disease. Medical therapy with anti-helmintic agents (most used Albendazole) by itself, is indicated in cases where surgical intervention is not possible for any reason, after multiple relapses and in the alveolar form of the disease. More often, it is used as a neoadjuvant or adjuvant therapy in combination with interventional and surgical procedures [11,12]. The most common and well-known treatment of LH is surgery. It has been a common pathology for years in the general surgical departments, especially in endemic regions. Surgical strategies can range from liver -sparing methods such as end cystectomy to more radical methods such as partial or total peri cystectomy and various hepatectomies. Immediate and late results of open surgery are very good. Recurrence rate after 1, 5 and 10 years was 100 %, 90.9 % and 87.9 %, respectively [7,13]. However, open surgery is associated with significant morbidity (15-25%) and mortality rate (up to 6,5%) and long hospital stay [7]. Newer minimally invasive methods of treatment such as laparoscopic and robotic surgery have the advantage of less morbidity, low cost, and shorter hospital stay [8,14]. Recent studies showed that laparoscopic treatment of liver hydatidosis is safe and effective in individual patients and has lower complications and recurrence rates. However, laparoscopy for liver hydatidosis could be very complex and challenging procedure in the cases of centrally located disease and suspected biliary complications. In other cases, laparoscopic approach can be a useful alternative to open surgery [7,13,14].

Although concept of management of liver hydatidosis is changing and going to less -invasive procedure, open surgery is still gold standard for complete cure in the complicated cases [15,16]. Nonoperative, percutaneous treatment of liver hydatidosis (PAIR) was introduced in the mid-1980s [17,18]. Initially received with skepticism by some, it has developed into an attractive alternative to surgery and medical therapy. In this treatment modality, the aim is to destroy the germinal layer with scolicidal agents or to evacuate the entire endocast. From the point of view of diagnosis, PAIR proved to be the only method that enables direct diagnosis of the parasitic nature of the cyst [11]. The most commonly used scolicidal reagents are hypertonic (20-30%) NaCl solution and 95% ethanol. Percutaneous cyst drainage has been shown to be an effective and safe procedure, with a low complication rate. The most serious complication of the PAIR method is chemical cholangitis, which is caused by the penetration of scolicidal agents into the biliary tree. Therefore, special attention is paid to the assessment of possible communication between the biliary cyst and the biliary tree (CBC). In addition to per-forming ERCP or MRCP, prior to cystic aspiration, contrast material is injected, which enables direct visualization of the communication with the biliary ducts [11,19,20]. Current guidelines give the indication for percutaneous treatment of hydatid liver disease. According to these guidelines, the best results with PAIR achieved in >5 cm CE1 and CE3a cysts (Table 1). Indications for this technique are uncomplicated cystic, multiple cysts if accessible to puncture, infected cysts, patients who refuse surgery, patients who relapse after surgery, patients in whom surgery is contraindicated. However, PAIR is contraindicated for CE2, CE3b, CE4, and CE5 cysts, early pregnancy, as well as for lung cysts, superficially localized cysts, cysts that communicate with the biliary tree [11,19,20]. In our case, there weren't any kind of contraindications of this procedure.

After PAIR had been performed, the patient had no problem after the procedure, he was discharged from the hospital. However, he was readmitted to University hospital with obstructive jaundice and cholangitis, one month after the procedure. All the laboratory findings, and MRCP showed biliary obstruction and cholangitis caused by scolex debris trough obvious CBC. CBC could be primary and secondary. Primary CBC are results of spontaneous post-necrotic cyst rupture into the biliary tree. It presents the most common complication of liver hydatid disease, with an incidence of 10%-37% for occult rupture and 3%-17% for frank rupture [14]. History of cholangitis, high bilirubin, high ALP levels, a cyst larger than 10 cm, central localization and the presence of suggestive US findings are clinical predictors of CBC. In these situations, periinterventional MRCP or ERCP are mandatory to detect biliary complications and perform sphincterotomy to decrease the pressure in the biliary tract, [21]. In our case, CBC was not suspected on the basis on pretreatment analysis of clinical signs, laboratory and imaging. Secondary (post treatment) biliary fistula occurs like a complication of the percutaneous treatment in 1.7%-6.2% of cases [19,21-23], which is comparable to 3,7% postoperative CBC after open surgery [7]. However, CBC complicated with biliary obstruction is extremely rare condition. The exact frequency of cholangiohydatidosis with mechanical

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icterus and secondary cholangitis is unclear. There are only 15 reports about cholangiohydatidosis, and secondary cholangitis founded by Manterola et al. [9].

To the best of our knowledge, our report is the first case report of mechanical icterus resulting as a late complication of the PAIR treatment of liver hydatidosis. The question of real etiology of post interventional CBC arises is it really caused by an iatrogenic injury to the biliary system with a needle or catheter, or it was actually silent and unrecognized during the intervention. It is a well-known fact that small CBC becomes visible only after any procedure (surgical or percutaneous) leading to lowering the pressure in the cystic cavity. Such CBC could be recognized during PAIR, based on the appearance of the yellowish biliru-bin-rich aspirate, which analysis is an integral part of the PAIR protocol. In this situation, the contrast instillation into the cavity could recognize silent and small CBC and confirm the diagnosis. In this case, no biochemical analysis of aspirate nor contrast imaging was performed during this PAIR procedure. As a consequence of the previous mistakes, a final mistake was made - the instillation of sclerosing agents into the cystic cavity and through the CBC into the biliary system. This could possibly have led to chemical cholangitis. Fortunately, cholangitis was localized and resulted in enlargement of the CBC itself. Necrotic debris migrated trough widen CBC and obstructed the biliary tree in the late period, one month after the intervention. Moreover, aspiration and extraction of parts of the cyst and necrotic debris was not complete, which indicated placement of the catheter in the cystic cavity.

On the 7th day after the PAIR procedure, the previously placed drainage catheter was extracted, without the complete visualization of the cystic cavity. No matter what, communication between hydatide cyst and right bile duct have resulted by protosco-lex entrance and bile duct obstruction, like in our patient. The complications seen in our case were accurately diagnosed and treated in our hospital. The goals of treatment of cholangiohydatidosis and secondary cholangitis is to make desobstruction, lavage and to drain the bile duct, as well as eliminate the source of the parasitic material. This can be performed by endoscopic or surgical manner (open or laparoscopic). The first choice in treatment could be ERCP with sphincterotomy, debris and scolex extraction and lavage of the biliary tract [11,15,16]. Despite the advantages of minimally invasive surgery, endoscopy and laparoscopy does not allow rational and definitive treatment of the whole problem.

Therefore, surgery remains the option of choice resulting with the most exacted biliary clearance and drainage (using T tube, combined external-internal or internal biliary drainage by biliodigestive anastomosis). The most significant advantage of open surgery is possibility to deal with the primary etiology -performing CBC closure and simultaneous resection of the hydatid cyst [15,16,22,23]. Detailed steps of surgical treatment of CBC and hydatide biliary obstruction depends on the site and size of the fistula [11,24]. Open technique, careful inspection, using of the intraoperative ultrasound and intraoperative cholangiography were specific procedures which confirmed the site and size of the cysto-biliary communication [25]. After careful lavage of the biliary tree, and correct intraoperative cholangiography, external-internal biliary drainage was successfully performed in our case. Careful analysis of certain available methods to cure complicated CBC is lacking. However, the actual number of reported cases is insufficient to conduct a comparative study with a methodologically adequate statistical power and level of confidence. Because of that, open surgery remains the most complete and powerful method of treatment [9].

Conclusion

Without timely diagnosis and management, complications of liver echinococcosis which occur in about one-third of patients may be life-threatening. This case report suggest that intra-biliary rupture and scolex obstruction might be a complicated natural course of necrotic changes in hydatide cyst and biliary tree and missed during PAIR procedure, either occurred iatrogenic ally during the implementation of a therapeutic procedure. CBC requires an early diagnosis, and a decision on further treatment, for a better prognosis of the disease. Surgery, however, is the gold standard in the treatment of hydatid liver disease, as well as in the treatment of its complications.

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