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Case Report

Percutaneous Treatment of Atypical Recurrent Subcapsular Hydatid Cysts after Surgery and Subcapsular Liver Hydatid Cysts Tow Case Report

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Abstract

Echinococcosis, or hydatid disease, is caused by the larval forms of taeniid cestodes belonging to the genus Echinococcus. Echinococcus granulosus and E. multilocularis are the primary species responsible for human echinococcosis, and mostly they affect the liver. The disease course is typically slow, and the patients tend to remain asymptomatic for many years. is a serious disease, potentially lethal, which can be found anywhere in the world, but especially in endemic areas such as the Mediterranean Basin, Australia, New Zealand, North Africa, Eastern Europe, the Balkans, Middle East and South America. The hydatic cyst is mainly found in the liver (75% of the cases), being asymptomatic in most cases and discovered accidentally on a routine abdominal ultrasound or an ultrasound performed for diagnosing other pathologies. The hepatic hydatid cyst therapy is multimodal, including medical, surgical, and, lately, minimally invasive techniques.

Keywords: Hydatid cyst, Recurrent hydatidosis, Subcapsular Hydatid cyst, percutaneous drainage, pair technique

Introduction

Echinococcosis, a group of zoonoses caused by cestodes of the genus *Echinococcus*, is one of currently twenty neglected tropical diseases (NTDs) [1]. With an estimated loss of over one million Disability Adjusted Life Years (DALY's) annually worldwide, it imposes a high burden on health [2, 3]. Parasitologically, an infected human is a dead-end intermediate host, disrupting the life cycle. Clinically, human echinococcosis presents predominantly in three forms, each requiring a different therapeutic approach. The fox tapeworm *Echinococcus multilocularis* causes the alveolar type, also endemic to Western Europe [4].

Many hydatid cysts remain asymptomatic, even into advanced age. Data from alveolar echinococcosis registries in Europe show that more than half of the patients are asymptomatic at diagnosis (56.2% in the FrancEchino registry in 2011-2021). [5]

Vague abdominal (right upper quadrant) pain is the most common presenting symptom (30%) and can last for years before lesions develop.

Jaundice is the most common presenting symptom, especially China. Progressive gradual cancerlike onset of jaundice is observed in most cases that involve symptomatic cholestasis.

Intermittent jaundice may also be associated with acute right upper quadrant pain when parasitic material migrates through the common bile duct. Hepatomegaly may be observed.

In the presence of bacterial superinfection, fever and chills may accompany gallstonelike symptoms. Fever and chills may also evoke liver abscess due to superinfection in the central periparasitic necrosis. 'Liver abscess-like' acute pain with or without fever may also reveal rapid growth of the lesions in the liver of immunocompromised patients. (6,7)

Various symptoms, ranging from dyspnea and bile sputum to seizures and stroke, as well as bone pain or skin tumor, may be the presenting symptoms of a secondary location or metastasis of the parasitic lesions (approximately 10% of cases).

The diagnosis of cystic echinococcus is mainly made on the basis of clinical presentation and imaging and serologic studies.

Despite the availability of various therapeutic options and the inherent risks (medical, percutaneous or surgical treatments), surgery remains the most frequently performed treatment, particularly in complicated or recurrent cases (8,9). One of the primary challenges in managing CE is postoperative recurrence, defined as the appearance of new active cysts after surgical intervention. Its estimated incidence is 8% (95% confidence interval [95% CI] 6–10%) (10), and has been associated with factors, such as the presence of multiple cysts (11,12), larger cyst diameter [11, 13], evolutionary complications [12, 13], and the type of surgical procedure performed [14,15,16]. In addition, perioperative benzimidazole therapy is consistently recommended in clinical guidelines as a preventive measure; however, the certainty of evidence remains low and consistent effect estimates are lacking (17,18,19).

Percutaneous drainage PAIR described by Ben- Amour et al. (20,21). PAIR means the abbreviation of puncture, aspiration of cyst content, injection of hypertonic saline solution or ethanol 75%-95% and reaspiration of all fluid after 5 to 15 minutes. PAIR technique may be carried out by either only US guidance or US and Flouroscopy guidance in combination. PAIR is the preferred treatment for WHO-IWGE type CE1 and CE3a hepatic cysts.

Main indication for percutaneous treatment of liver CE cysts includes viable CE types such as CE1, CE2, CE3a and CE3b according to WHO classification.There is a direct relation between the type of liver CE

cysts and the percutaneous technique which will be employed. Therefore, CE1 and CE3a are treated by either PAIR or Catheterization techniques while CE2 and CE3b are treated by MoCaT technique.

The infected hydatid cysts are treated by percutaneous drainage and antibiotics like other liver abscesses (22). Recurrent hydatid cysts are also treated by percutaneous techniques (23). Suspected postoperative fluid collections are also treated bypercutaneous approach

Percutaneous treatment in the ruptured liver hydatid cysts into biliary system may be performed together with endoscopic interventions. In this case,a catheter is placed into the cavity of the ruptured cyst to the biliary system as a first step. On the second step, the patient is referred to gastroenterology for endoscopic intervention in order to clean main bile duct by a balloon catheter during ERCP after papillotomy.

The procedure is ended up with insertion of plastic stents into the main bile duct. By both intervention fistulae between CE cyst cavity and biliary system ceased in a short period of time. Surgery is and alter-

native option for these cases (24,25). The size, number or localization of the hydatid cysts in the liver are not deemed to be contraindications for percutaneous treatment. Any treatment is unnecessary in patients with CE4 and CE5 liver hydatid cysts as these types are managed by 'Wait and Watch approach (26) .

We report Two cases of hydatid infections have been documented an unusual clinical presentation of recurrent hydatid cyst of liver. A 33-year-old female patient presented to us with complaints of pain in abdomen for months. The patient gave history of being previously operated for hydatid cyst of liver 6 years back. His abdominal radiograph and computed tomography scan revealed a cystic lesion in the right lobe of liver. And a 44-year-old woman reported experiencing intermittent abdominal pain and discomfort over the past five months, primarily centered in the right upper quadrant his abdominal radiograph and computed tomography scan a subcapsular hydatid cyst with no signs of cyst rupture or secondary infection. The two patients managed by percutaneous drainage the so-called PAIR (**P**uncture, **A**spiration, **I**nstillation, and **R**easpiration) technique and received chemotherapy for 6 months and the patient had a good recovery.

Case Report

Case 1 A 33-year-old female patient presented to my clinic complains of pain in abdomen for months. She was a housewife in a rural area

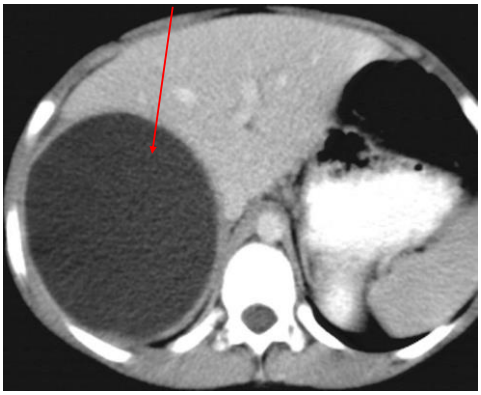
The patient living in rural area containing cats and dogs.

She also stated that she had undergone surgery to remove a hydatid cyst from her liver six years prior.

On clinical examination, there was a scar in right subcostal region. A lump was felt in the right hypochondrium, which was tender on palpation. His ultrasonography abdomen revealed a hydatid cyst in right lobe of liver (Fig.A 1)Computed Tomography (CT) scan of abdomen and chest was done which helped to exclude cases of tumors and other benign cysts. It showed a fairly well-defined rounded lesion with septae and multiple cysts in right lobe of liver. Size of the cyst was 13.5 × 11.0 cm. CT scan of chest showed no cystic lesions (Fig.A 2). His haemoglobin percentage was 13.2 gm/dl and total leucocyte count was 9.6 × 10^9/L with no eosinophila. Liver function tests showed Total bilirubin was 0.8mg/dl with direct bilirubin 0.4 mg/dl and indirect bilirubin 0.4 mg/dl. SGOT (46 IU/L), SGPT (25 IU/L) and Alkaline Phosphatase (86 IU/L) levels were normal. Following a radiological finding and past history of operative hydatid cyst the diagnosis recurrent of hydatid cyst was made and the patient was planned for hepatic cyst.

(Fig.A 1) A-Abdomen ultrasound showing lesion in right lobe of liver

(Fig. A2) CT abdomen showing lesion in right lobe of liver



Informed consent was taken from the patient for percutaneous drainage called PAIR (**P**uncture, **A**spiration, **I**nstillation, and **R**easpiration) technique. The patients are given Albendazole (10 –15 mg/kg/day) one week before the procedure for the aim of prophylaxis in order to reduce the risk of the abdominal dissemination [31]. patients fast overnight for approx. 8 h before the procedure Done under US ,Under local anaesthesia cyst is **punctured as first step** using a spinal needle 18 gauge , second step CE cyst content is aspirated as much as possible. The amount which aspirated 350 ml then the third step injected the scolicidal agent as ethanol (90%) into the cyst cavity. The volume of these agents should be 30% of aspirated volume, ethanol should be kept within the cavity at 15 min is needed to follow separation of endocyst from adventitia (pericyst) which generally occurs within 15 min, then a last step respiration of the cyst fluid and after respiration, an ultrasound examination of the cyst is done to confirm complete detachment of the laminated membrane from the cyst wall. Before needle withdrawal, normal saline is installed into the cyst cavity. the needle is withdrawn after respiration of all the cyst content, Procedure success is defined if the laminated membranes are detached in toto, and the scolex viability test shows the scoleces in the reaspirated fluid are nonmotile on light microscopy and take up the color on vital dye staining. As figure (A 3 a,b)

figure (A 3 a)

Aspirated fluid of hydatid cyst

figure (A 3 b)

Aspirated fluid of hydatid cyst

After ethanol injection

figure (A 3a, b)

A routine abdominal ultrasound examination is performed within 24 h to observe cyst appearances and rule out any cyst leak, and if uneventful, the patient was discharged on a regime including Albendazole (10mg/kg/day) for 6 months, as it was a case with recurrence. He wase regular follow up from last 12months to the clinic. After three years there is no further recurrence and the patient has shown good recovery as figure (A 4)

figure (A 4)
US shown good recovery

Case 2 A 44-year-old woman reported experiencing intermittent abdominal pain and discomfort over the past five months, primarily centered in the right upper quadrant. Her medical history is notable for its lack of significant issues or known allergies. During the physical examination, mild tenderness was observed in the right upper quadrant, though she showed no signs of acute distress or rebound tenderness, indicating a non-urgent condition.
Ultrasound: Revealed a subcapsular hydatid cyst in the right liver lobe, approximately 6 cm in diameter. The cyst appeared to be near the liver capsule, raising concerns about the risk of rupture (Figure B 1).

CT scan: Confirmed the presence of a subcapsular hydatid cyst with no signs of cyst rupture or secondary infection. The liver is mildly enlarged, measuring 17 cm in the cranio-caudal direction. The parenchymal texture is homogeneous. A large cystic mass is seen in segments V and VI of the liver, measuring approximately 57.6×42.2 ×38.7 mm (Figure B 2). The lesion shows peripheral calcifications but no perifocal reaction. The interface between the liver and the suprarenal gland is normal. The portal venous system is normal in calibre, and intra-hepatic biliary radicles are not dilated. The porta hepatis is free of lymph nodes.

Figure B 1

Ultrasound: Revealed a subcapsular hydatid cyst in the right liver lobe, approximately 6 cm in diameter

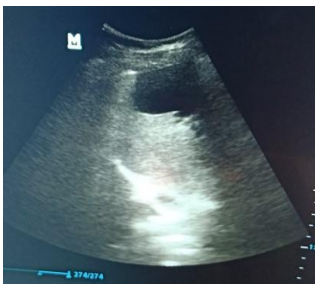
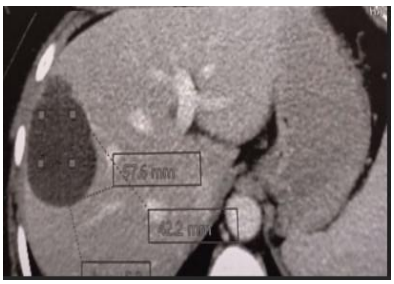


Figure B 2

CT scan: Confirmed the presence of a subcapsular hydatid cyst with no signs of cyst rupture or secondary infection



USG-guided catheter drainage of a right lobe hydatid cyst. The procedure involved insertion of an 18G LP needle into the cyst, with subsequent placement of an 18 Fr. The cyst contents were aspirated. Absolute alcohol was used as a scolicidal agent as ethanol (90%) to sterilize the cavity, followed by reaspiration after 15 minutes and repeated injection of ethanol and followed by reaspiration after 15 minutes for complete evacuation (Figure B 3).

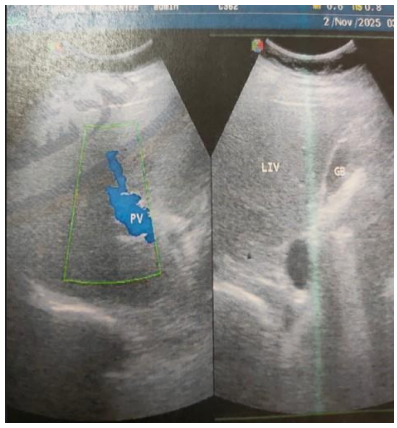
Sand samples taken pre- and post-procedure showed no motility, indicating successful treatment. The procedure was performed under aseptic conditions and was uneventful.
Follow-up imaging (ultrasound and CT) at 3- and 6-months post-procedure showed significant reduction in cyst size with no evidence of residual disease or complications.

Figure B 3 a
Aspirated cystic fluid

Figure B 3 b
Aspirated cystic fluid
after ethanol injectipn

Follow up after 12 months there is no further recurrence and the patient has shown good recovery as figure (B 4)

figure (B 4)
US shown good recovery



Discussion

Hydatid disease, a life-threatening parasitic zoonosis, is estimated to affect 2-3 million people worldwide. It is most prevalent in endemic regions such as the Mediterranean Basin, Eastern Europe, North Africa, the Middle East, South America, Australia, and New Zealand, with incidence rates reaching up to 50 cases per 100,000 person-years. In highly endemic areas, the prevalence can be as high as 10% of the population. The disease is caused by the larvae of Echinococcus granulosus and Echinococcus multilocularis and is transmitted via the fecal-oral route, often through close contact between dogs, sheep, and humans. Dogs serve as intermediate hosts in this transmission cycle. Around 75% of hydatid disease cases involve the liver, with approximately 80% affecting the right hepatic lobe and 20% the left lobe. The lungs are the second most commonly affected organ, accounting for about 15% of cases. Other organs such as the kidneys, spleen, peritoneal cavity, skin, and muscles are less frequently impacted. (27,28) .

There is a controversy regarding the role of fine needle aspiration cytology (FNAC) in diagnosis of hydatid cyst. The risks are chances of rupture of the cyst, anaphylaxis and dissemination (29). Ultrasonography, computed tomography, and magnetic resonance imaging (MRI) are superior to plain radiography in diagnosis of hydatid cyst (30). CT scan with contrast may demonstrate a thin enhancing rim if the cyst is intact (31). In the present case, the diagnosis was made by typical appearance on computed tomography.

Surgery is considered as the standard treatment for HHC. However, surgery is not without risks and there is a high incidence of dissemination, infection and recurrence of 2% to 25%, with morbidity of 0.5% to 4% (32,33). Furthermore, surgery is not advisable in elderly patients with cardiac or pulmonary disease, nor in recurrent cases. Medical treatment alone in the form of mebendazole, and recently albendazole and praziquantel, have been used as an alternative therapy to surgery, but the success rate in terms of a reduction in size of HHC and the change in echotexture has been variable (34,35).

When managing individuals who are not candidates for surgery and

have many cysts or multiorgan involvement, these anthelmintic drugs can be administered as monotherapy for early-stage cysts (CE1, CE3a) (36,37) . Randomized Controlled Trials (RCTs) have demonstrated that albendazole is more effective than mebendazole, with superior outcomes in terms of cyst degradation and cure rates (38).

Percutaneous techniques, considered minimally invasive procedures, offer an alternative to chemotherapy and surgery. These techniques include PAIR (Puncture, Aspiration, Injection, and Re-aspiration), PAIR-D, the Modified Catheterization Technique (MoCaT), and Percutaneous Evacuation (PEVAC). Among these, PAIR is widely recognized as superior to catheterization. In the PAIR procedure, cysts are first identified using ultrasound guidance. A local anesthetic is then applied to puncture the cyst percutaneously. Once the cystic fluid is aspirated, scolicidal agents such as alcohol, betadine, cetrimide, or hypertonic saline are injected into the cyst cavity. After 20 to 30 minutes (39), the injected solution is re-aspirated, completing the procedure. PAIR-D, a variation of PAIR, involves inserting an intracystic catheter after the initial steps. The cavity is then emptied and irrigated with saline solution after 24 hours. PAIR has gained prominence as an effective, cost-efficient procedure with a high success rate, particularly for CE1 and CE3a cysts (39,40,41). However, its use with CE2 cysts remains a subject of debate. It is recommended for patients who are unsuitable for surgery due to contraindications, those experiencing post-surgical relapses, patients refusing surgery, and those with multiple accessible cysts. It is also effective for hydatid cysts containing daughter vesicles, detached membranes, or superinfection, as well as use in pregnant women or individuals unresponsive to medication therapy. However, it is contraindicated in uncooperative patients, those with inactive or calcified cysts, cysts that cannot be punctured, or cases where there is communication with the biliary tree. To mitigate the risk of subsequent hydatidosis, a combination of serologic tests and imaging should be carefully monitored. PAIR is particularly recommended for surgical relapses, inoperable cases, or as a first-line treatment for CE1 and CE3a cysts alongside albendazole therapy for prophylaxis. Many studies have concluded that PAIR combined with albendazole provides better outcomes than surgery and is considered the first-choice treatment for uncomplicated hydatid cysts ((42,43). Despite concerns surrounding cysts in subcapsular locations, successful outcomes with PAIR have been demonstrated. Critical factors contributing to success include appropriate premedication to prevent anaphylaxis, careful ultrasound-guided puncture, and the use of scolicidal agents to sterilize the cyst cavity. Ensuring no biliary communication through contrast injection further enhances safety. Follow-up observations have shown significant cyst size reduction and a lack of complications, reinforcing the efficacy and safety of PAIR in even unconventional cases.

Recurrence of hydatid cyst is defined as the appearance of new and growing hydatid cysts after therapy. It includes reappearance and growth at the site of previously treated hydatid cyst or the appearance at a new distant site due to spillage (44,45). Our patient also had history of previous operation for hepatic hydatid cyst 6 years back. He had recurrence at site of previously treated cyst. Two most important causes for recurrence are minute spillage of the hydatid cyst and inadequate treatment due to missing cysts and incomplete pericystectomy (46).

The PAIR procedure is an effective method for managing hydatid cysts; however, its application in subcapsular cysts and large recurrent cysts carries significant risk due to the possibility of rupture and severe allergic reactions. These cases demonstrate that, with meticulous patient selection, adequate premedication, and rigorous monitoring, PAIR can be safely utilized even for subcapsular hydatid cysts and recurrent. The positive outcome observed in this patient indicates that, although generally contraindicated, PAIR might be a viable option in particular cases when proper precautions are in place.

Conclusion

Hydatid disease presents with a range of clinical manifestations. A high index of suspicion warrants prompt radiological evaluation, as chest radiographs (CXR), ultrasonography, and CT scans are effective in diagnosing the majority of cases. In suspected scenarios, both thoracic and abdominal regions should be assessed due to the relatively frequent coexistence of the condition in these sites.

These cases report highlights that, contrary to traditional guidelines, the PAIR procedure can be effectively and safely utilized for managing subcapsular hydatid cysts and recurrent under carefully controlled conditions. Additional studies and cases reports are necessary to further refine the indications and protocols for employing PAIR in similarly complex scenarios.

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Authors' contributions

All authors are responsible for the modification and giving final approval of the manuscript. Abdelmoneim Elhadidy was a contributor in writing the manuscript. All authors read and approved the final manuscript.

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Availability of data and materials

Please contact the corresponding author for data requests.

Declarations

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Consent for publication

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Competing interests

The authors declare that they have no competing interests.

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