Challenges in Management of Hydatid Cyst

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Introduction

Hydatid cyst disease is a parasitic infection in which humans are accidentally affected by ingesting larval forms of Echinococcosis granulosus. The common sites of hydatid cyst are liver, lung and spleen. Unusual sites of hydatid cyst reported in this report includes peritoneum.

Case Reports

Case 1

A 35-year-old male presented to the outpatient department with dull aching right hypochondrial pain for 10 days, which was associated with fever & chills. On examination, the patient was febrile, icteric with tender hepatomegaly. Total leukocyte count and liver function test were raised. Serum bilirubin was elevated. USG abdomen showed 5x6x6cm, heterogenous, hypoechoic lesion with internal cystic areas with peripheral calcification in right lobe. CECT abdomen (Figure 1a) showed daughter vesicles and MRI was suggestive of hydatid cyst. Patient underwent laparotomy wherein calcified hard cyst in the 5th & 6th segment of right lobe was found. Cyst fluid and daughter cysts were evacuated (Figure 1b) and 5% povidone iodine was instilled into cyst cavity for about 10min. Capitonnage was done by obliterating cyst space with vascularised omentum. Post op was uneventful. Patient was followed up for six months and it was uneventful.

Figure 1a: CECT abdomen.
**Case 2**

A 50 years old female came with complaint of abdomen pain for the past 4 months. Clinical findings were unremarkable. USG abdomen showed multiloculated cystic lesions in the right lobe of liver and CECT abdomen showed cystic lesions with daughter cysts in segment 8 of right lobe of liver with calcified wall (Figure 2a). Blood parameters were unremarkable. Patient underwent laparoscopic surgery. Intraoperatively, cyst wall identified in right lobe of liver (Figure 2b) Using verres cyst wall was punctured and aspirated. 3% Nacl was injected. Partial pericystectomy was done using harmonic scalpel. Cyst cavity capionnaged & obliterated with omentum. Post op was uneventful. Patient was prescribed Albendazole with praziquantel and was followed up for a year.

**Case 3**

A 58-year-old female presented with abdomen pain and distension for 3 months which was sudden in onset and rapidly progressive in nature with loss of weight and appetite. Patient had past history of thoracotomy done for cystic lesion of the left lung 7 years ago and diaphragmatic rent was created during the surgery. Blood parameters were unremarkable. X-ray lung showed cystic lesions of the left lung and CECT abdomen revealed large cystic lesions in the left hypochondrium and left iliac fossa. CECT thorax showed multiple cystic lesions involving the upper lobe of left lung (Figure 3a) Patient posted for laparotomy and intraoperatively a large cyst containing 2 litres of fluid was found in the left hypochondrium adherent with the transverse mesocolon (Figure 3b). Multiple cystic and calcified lesions were found in the falciform ligament, left hemidiaphragm and peritoneum. All of them were excised and sent for histopathological examination. Pre and postoperatively she was started on albendazole 15mg/kg/day for a period of 21 days.
Case 4

A 29 years old female came for removal of intrauterine device. On per abdominal examinations she had vague lower abdomen fullness and per vaginum examination revealed missing thread. Further, on investigation Cect abdomen revealed incidentally multiple cystic lesions were noted in the lesser sac, root of mesentry abutting Duodenal jejunal flexure and right side of the pelvic cavity and also in the right lobe of liver 6th segment (Figures 4a&4b). Pre operatively, patient was prescribed Albendazole for 14 days. Intraoperatively, three calcified cysts were found in the lesser sac, liver, pelvis displacing the uterus to left and Cyst in lesser sac was removed in to (Figures 4c&4d). Partial pericystectomy was done for liver and pelvic cyst. Post op was uneventful and was prescribed Albendazole for six months with regular follow up.
Case 5

40 yrs old female presented with symptoms of gastritis. Incidentally, a 6x5x5cm cyst was noted in spleen in sonogram which was suggestive of hydatid cyst. Blood parameters were unremarkable. Cect abdomen showed a large cyst in the spleen with calcification. Patient underwent laparotomy and Intraoperatively, a large calcified cyst was found in the spleen. Splenectomy was done. Pre and postoperatively she was started on albendazole 15mg/kg/day for a period of 21 days and followed up for six months.

Discussion

Hydatid cysts are located mainly in the liver (75%), lung (15%) and to lesser extent in the bones, kidney, spleen, muscles, central nervous system, eyes and serous surface like pleura or peritoneum which provides friendly micro environment for development of the cyst (Prousalidis J, et al.1998) [1].
Hydatid cyst most commonly present in right lobe (78.2%), left lobe (13.4%) and in both lobes (8.2%). Most common clinical presentation is abdominal pain (79.06%) and as abdominal mass (20.93%). The Indirect Hemagglutination (IHA) and ELISA is associated with diagnostic sensitivity rates up to 85%-96% (Lorenzo C, et al.2005) [2]. E. granulosus antigen B and antigen 5 are the most specific antigens used for immunological diagnosis. However immunological methods show cross reactivity with other parasitic antigens or with non-parasitic diseases such as malignancy or liver cirrhosis (Rinaldi F, et al.2014) [3]. Percutaneous Fine Needle Aspiration (FNA) biopsy ultrasound guidance is done in suspected case. Presence of protoscolexes, cyst membrane or Echinococcal antigen or DNA in aspirated fluid confirms the diagnosis. However, the risk of anaphylaxis is 2.5% (Neumayr A, et al. 2011) [4].

Ultrasound is gold standard screening test for hydatid cyst. In case of emergency presentation and screening for multi organ involvement computed tomography is used (Keong B, et al.2018) [5]. 55% of the cases are diagnosed with help of computed tomography and ultrasound. Ultrasound alone detects 36% and computed tomography alone 7% of the cases reported on hydatid cyst. MRI is performed as second line imaging and better demonstrate biliary involvement. Anti-hydatid Ig G4 ELISA was used for diagnosis and to detect early recurrence. Increase in Ig G4 antibodies were detected in patients which gradually decreased in intact univesicular and multivesicular cyst with few larger daughter cysts, cyst with diameter of under 6 cm. 99.5% technical success rate was obtained using PAIR (Puncture, aspiration of cyst content, injection of hypertonic saline solution and reaspiration of all fluids) treatment. This is sufficient to decrease recurrence rate. Combination of Albendazole and praziquantel for a period of 2-6 months lead to complete disappearance of 47.4% of cyst whereas when Albendazole was administered for period of 6 months to 2 year lead to disappearance of 36.84% of cyst. Hence combination of Albendazole and praziquantel is preferred over Albendazole as it is more effective and takes shorter duration (Mohamed AE, et al.1998) [10].

Gharbi classification of hydatid cyst is used for characterizing the cyst which is done with the help of ultrasound. Hence, ultrasound is the primary imaging modality.

Types of cyst according to Gharbi classification (Gharbi HA, et al. 1981) [7]:

- **Type I**: Pure fluid collection
- **Type II**: Fluid collection with a split wall (Water Lily sign)
- **Type III**: Fluid collection with septa (Honeycomb sign)
- **Type IV**: Heterogenous echographic pattern
- **Type V**: Reflecting thick wall

Gharbi classification were improved as World Health Organisation (WHO) in 2001 (WHO Informal Working Group; 2003) [8].

WHO stage. Characteristics. Activity

- **CE**: Unilocular, anechoi cyst with double lime sign Inactive
- **CE2**: Multiseptated rosette sign, honeycomb pattern Inactive
- **CE3a**: Cyst with detached membrane (Water lily sign). Transitional
- **CE3b**: Daughter cysts in solid margin Transitional
- **CE4**: Heterogeneous cyst, no daughter vesicles. Inactive
- **CE5**: Solid margin with calcified wall. Inactive

Magnetic Resonance Imaging (MRI) can demonstrate all the features of hydatid disease, with exception of calcification and it is used in monitoring the response to treatment.

Clinical success rate of using Albendazole medication as an adjuvant to percutaneous treatment was 96.1%. Side effects of using Albendazole were reported in 24.1% (Akhan O, et al.2014) [9]. Albendazole should be administered a week before and one month after PAIR (Puncture, aspiration of cyst content, injection of hypertonic saline solution and reaspiration of all fluids) treatment. This is sufficient to decrease recurrence rate. Combination of Albendazole and praziquantel for a period of 2-6 months lead to complete disappearance of 47.4% of cyst whereas when Albendazole was administered for period of 6 months to 2 year lead to disappearance of 36.84% of cyst. Hence combination of Albendazole and praziquantel is preferred over Albendazole as it is more effective and takes shorter duration (Mohamed AE, et al.1998) [10].

Gomez, et al. revealed that the recurrences is observed more frequently in patients undergone conservative procedure compared to radical surgery (24% vs. 3%), and the morbidity was detected to be higher (11% vs. 3%) (Gomez I, et al.2015) [11]. Surgical procedure includes tube drainage, capitonnage, omentoplasty, segmentectomy and cystoenterostomy. Tube drainage causes infection (11.84%), biliary fistula (3.2%), cholangitis (1.9%). Capitonnage caused cholangitis (2.3%) and infection (0.9%) (Balik AA, et al.1999) [12]. Isolated omentoplasty filling technique is a safe management in filling the residual cavity after surgery with less operative time because omentum has high absorptive capacity and capacity to fill the residual cavity. Omentoplasty can be combined with pedicle omental pack or with isolated omental pack. Pedicle omentoplasty technique produce similar result compared to isolated omentoplasty (Aldelaocoy A, et al.2016) [13].

Percutaneous drainage is preferred over surgical management of hydatid cyst as there is less hospital stay, more complete disappearance of cyst (88% vs 72%) and less complication (32% vs 84%). Percutaneous treatment is an effective and safe method for treatment of uncomplicated hydatid cyst where mean volume reduction of 77.5 % is seen (Kahrman G, et al.2017) [14]. PAIR (Puncture, aspiration of cyst content, injection of hypertonic saline solution and reaspiration of all fluids) is highly effective method in intact univesicular and multivesicular cyst with few larger daughter cysts, cyst with diameter of under 6 cm. 99.5% technical success rate was obtained using PAIR (Bakdik S, et al.2018) [15]. Percutaneous puncture in second trimester of pregnancy is effective.
safe procedure for prevention of complications of echinococcus in late pregnancy and childbirth (Musaev GK, et al.2019) [16]. Complication rate of 6.3% was noted (Kahriman G, et al.2017) [14].

Alcohol as scolicidal and sclerosing agent during percutaneous treatment is associated with high success rate and low rate of recurrence and complication. Hypertonic saline is another potent scolicidal agent and its effectiveness varies with concentration and Time of contact. When 20% hypertonic saline was used for 5 mins, 50% of the scolex were alive and when left for 9 mins all the scolex were destroyed. 10% hypertonic saline left for 30 mins, 10% of scolex were alive and 5% hypertonic saline used for 30 mins, 70% scolex were alive. 10% povidone iodine, 0.5-1.5% Cetrimide, 10% sodium hypochlorite and 3% hydrogen peroxide are the common scolicidal agent used for treatment (Mustafa Karaoglanoglu.2004) [17].

Laparoscopic approach for the treatment of liver hydatid cyst was associated with higher rate of extra-hepatic and peritoneal recurrence than laparotomy. Common laparoscopic procedure includes cystectomy (60.39%) followed by partial pericystectomy (14.77%) and total pericystectomy (8.21%). Conversion to laparotomy occurred in 4.92% where the common cause was due to anatomical limitations. Most common complication encountered during laparoscopic treatment was bile leakage which counts to about 6.2% and post-operative recurrence rate of 1.09% (Tuxun T, et al.2014) [18]. Laparoscopic treatment is better than PAIR as it as better clinical cure (98.7% vs 97.5%), less surgery related complication (7.32% vs 3.43%) (Chen X, et al.2015) [19]. Follow up sonography is done three times a year for three years following surgery. Reduction in size and volume of the cyst, thickening and irregularity of the wall, decreased fluid content and solid appearances of the remnant at ultrasound were accepted as positive criteria for healing.

As a complication of hydatid cyst of liver, biliary communication occurs in right duct (55-60%) and left duct (25-30%). Treatment for communicating hepatic hydatid cyst is surgery but it poses significant risk in management. Hence, following balloon occlusion, sclerotherapy was performed using absolute alcohol with successful ablation of hydatid cyst and thereby resolution of the biliary dilatation (Josef Varro, et al.2011) [20]. Recently, relative new treatment involving percutaneous thermal ablation of germinal layer in the cyst by means of radio frequency ablation device is currently under research and study (Sharma D, et al.2009) [21].

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