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*Research Article*

# ABDOMINO-CUTANEOUS FISTULA FOLLOWING RECURRENT HYDATID LIVER DISEASE WITH ABDOMINAL WALL HYDATID CYST: A CASE REPORT AND REVIEW OF LITERATURE

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## INTRODUCTION

Hydatid disease or Echinococcosis is a widespread zoonotic parasitic disease caused by a tapeworm that continues to be a clinical and public health problem worldwide, especially in areas where animal husbandry and subsistence farming form an integral part of community life. Hydatid disease occurs primarily in sheep grazing areas of the world but is common worldwide because dog is a definitive host.[1]

## CASE PRESENTATION

A 50years old woman presented to surgery OPD with complaints of pain right upper abdomen since one month and swelling in the right upper abdomen of one weekduration. The swelling was insidious in onset, gradually progressive and size was comparable to that of a lemon at presentation. It was associated with serous discharge coming out of the swelling which was the rare feature of this case. There was no history of fever, loss of weight or jaundice. Patient gave history of contact with pets like sheep, dogs, goats, etc.Patient gave past history of surgery for hydatid disease twice; once in 2004 and once in 2017. According to history (records of surgery not available with the patient), open surgery was done for liver hydatid cyst in 2004 and laparoscopic cystectomy of hydatid cyst was done in 2017. Examination revealed a painless cystic swelling in the right hypochondrium measuring 5 × 4 cm, tender on palpation with clear serous discharge coming from the abdominal wall swelling.

Ultrasound and CECT abdomen were done which showed enlarged liver with large well defined bilobed complex cystic mass lesion measuring approximately 7.2×6.2×9.5 cm in segment 8 and 5 of right lobe with dehiscence of its wall seen along the anterior aspect resulting in an organised walled off collection in the subcapsular hepatic region and a fistulous tract measuring approximately 32×21 mm seen in anterior abdominal wall in right hypochondriac region with the tract cranially seen communicating with the organised sub-capsular hepatic collection. A well defined cystic lesion measuring 5.0×3.1 cm seen in left lobe of liver with peripheral wall calcification and air foci/fat density component within. Another small well defined cystic lesion measuring of size 14×12 mm seen in segment 6 of right lobe. A subcutaneous cystic swelling measuring 4.0×5.2 cm with intramuscular extension seen in the anterior abdominal wall in the region of right hypochondrium which communicates with the subcapsular hepatic collection through the fistulous tract.

Her haemoglobin was 10.4 gm/dl,total leukocyte count was 4200 with no eosinophilia. Her liver function tests were within normal limits; total bilirubin was 0.9 and total protein was 7.9gm preoperatively. Chest skiagram was normal. Following a positive hydatid serology and radiological findings, diagnosis of hydatid cyst was made and the patient was planned for surgery for both hepatic and abdominal wall cyst. Informed consent was taken from the patient for the operation and there were no ethical issues.

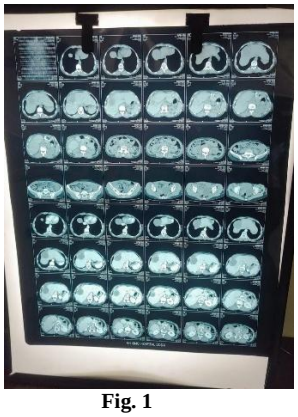

**Fig. 1**

**Fig. 2**

**Fig 1& 2** CT images showing hydatid cyst and connecting fistula

Tablet albendazole was started one week prior to surgery.Exploratory laparotomy with excision of abdominal wall cyst with excision of fistulous tract with partial cystectomy(liver hydatid)with cholecystectomy was done. Intraoperative findings were: (a)Fistulous tract present which was seen communicating with abdominal wall cyst and liver hydatid cyst in segment 5 and 8; (b)Dense adhesions were present between liver, GB and omentum. Packing of the abdomen was done with betadine(10% povidone iodine)soaked gauze to avoid anaphylaxis and seeding. Cyst

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was aspirated and bile tinged fluid came out, daughter cysts were removed, scolicidial agent(hypertonic saline) was injected and was aspirated and cystectomy was done,bile leak identified and was repaired,drain was kept in cavity and cholecystectomy was done.

**Fig. 3** Swelling on upper abdominal wall with fistlulous opening

Patient was started on oral sips on first postoperative day and tablet albendazole was started. There was minimal serous discharge from the drain for the next 5 days. Drain was removed on the 8th postoperative day. Wound was healthy and there were no complaints. Patient tolerated orals well and was ambulated early. Patient was discharged on the 11th postoperative day after suture removal. She was examined on follow up visits in the OPD at regular intervals for about one year and there were no complaints in the follow up period.

**Fig. 4**

**Fig. 5**

**Fig. 4** Fistulous tract connected to skin
**Fig. 5** Intra-operative picture of excision of the fistula

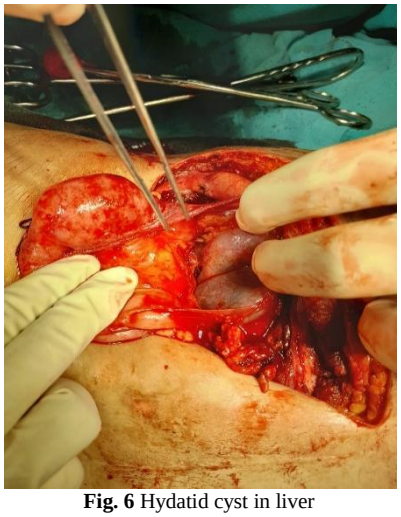

**Fig. 6** Hydatid cyst in liver

## DISCUSSION

In hydatid disease, humans contract the disease from dogs, but there is no human-to-human transmission. There are four species of echinococcus that cause hydatid disease. Echinococcus granulosus is the most common,and Echinococcus multilocularis, Echinococcus vogeli and Echinococcus oligarthus account for smaller number of cases.Dog is the definitive host of E. granulosus; sheep is the usual intermediate host. Humans are the accidental intermediate host and do not play a role in the biological cycle of the worm. Humans are an end-stage to the parasite. The mode of infection is by the ingestion of food contaminated with dog faeces and also by direct contact with dogs[2].

The most common sites affected are the liver (63%), the lungs(25%), followed by muscles(5%), bones(3%), kidney(2%), spleen(1%), and other sites(1%). Hydatid cyst of skeletal muscle is a rare condition accounting for 1–4% of all hydatid cyst disease[1.] Some cases of primary muscular hydatidosis at various sites have been reported, that is, biceps brachii[3], thoracic wall[4], sartorius[5,6], supraspinatus[7], gluteus[8], pterygoideus[9], and soleus muscles[10], whereas only few cases of primary subcutaneous hydatidosis have been reported[11].This low prevalence of abdominal wall hydatid cyst may be explained by the physical barriers to the hematogenous spread of the cysts which are present in hepatic sinusoids and pulmonary capillaries. There are various other factors which may make encystment of the parasite in these tissues less likely like muscle contraction and presence of high lactic acid in skeletal muscles, etc. Primary hydatid cyst of abdominal wall musculature is a very rare disease. Till date, only few cases have been reported as seen in this case report proving to be a diagnostic dilemma.[12] The ingestion of contaminated food leads to hatching of the ova in the gastrointestinal tract.

**Table 1** Literature review of the reported cases of abdominal wall hydatid cyst

| Author | Year | Age(year)/sex | Location | Any other Communication | Management |
|---|---|---|---|---|---|
| Abhishek et al[13] | 2012 | 60/ female | Right paraumbilical | No | Surgery |
| Gulmez et al[14] | 2015 | 60/ female | Left paraumbilical | No | Surgery |
| Tarahomi et al[15] | 2016 | 57 / female | Umbilical | No | Surgery |
| Our study | 2023 | 50/ female | Right hypochondrium | Communicating with liver hydatid cyst | Surgery |

The enclosed embryos are liberated in the gastrointestinal tract and transported to the liver by portal circulation. The liveracts as the first filter in trapping the embryos which then develop into hydatid cysts in 55–70% of cases, followed by the lungs as the second filter in 18–35% of cases. Some organisms escape from these filters and develop in other organs of the human body. The incubation period is highly variable. The cyst grows at a rate of 0.3–1 cm per year and may take 5–20 years to attain sufficient size to cause symptoms[3]. Three weeks post infection, a visible hydatid cyst develops which then grows slowly in a spherical manner. A pericyst or fibrous capsule derived from host tissues develops around the hydatid cyst. The cyst wall itself has two layers, an outer gelatinous membrane (ectocyst) and an inner germinal membrane (endocyst). In a definitive host, the scolices develop into an adult tapeworm; but in the intermediate host, they can only differentiate into a new hydatid cyst.

Hydatid cyst is diagnosed in equal numbers of men and women at an average age of about 45 years. Approximately 75% of hydatid cysts are located in right liver and are solitary. Most of the patients are asymptomatic until complications occur. The most common presenting symptoms are abdominal pain, dyspepsia and vomiting. The most frequent sign is hepatomegaly. Jaundice and fever are each present in approximately 8% of patients and rarely present as swelling on abdominal wall as in this case[2].

Rupture of the cyst into the biliary tree and bronchial tree or free rupture into the pleural, peritoneal or pericardial cavities can occur. Free ruptures can result in disseminated echinococcosis or a potentially fatal anaphylactic reaction[2].
Ultrasound is the most commonly used worldwide for the diagnosis of Echinococcosis because of its availability, affordability, and accuracy. A number of findings on ultrasound can be diagnostic but depend on the stage of the cyst. Calcification in the wall of the cyst is highly suggestive of hydatid cyst and can be helpful in the diagnosis. Similar findings are seen on CT scan or MRI scan. These cross-sectional imaging studies can also evaluate extra hepatic disease and demonstrate detailed hepatic anatomic relationships to the cyst[2].

Although the treatment of hepatic hydatid cyst is primarily surgical, alternative options are in evolution.There are various classification systems used for the staging of the hydatid cyst, most commonly being Gharbi's classification and WHO classification. After staging,the patient is managed accordingly.Surgical management is the basic treatment for hydatid disease.

Surgery for hydatid disease is divided into two subgroups-conservative and radical. Conservative surgery (evacuation of the cyst contents and partial peri-cystectomy) plus albendazole

achieves satisfactory long-term results. Radical surgery(total resection, cystopericystectomy) is preferred only in selected patients. Chemotherapy is used as a complement to the operative treatment to avoid recurrence[16]. Other treatment modalities include minimal invasive procedure like PAIR, modified PAIR, PEVAC, etc which can be done in selected cases.

In our case, conservative surgery was followed by chemotherapy. Albendazole is a broad-spectrum oral anti-helminthic drug which acts by blocking glucose uptake of the larvae and the adult worm. The glycogen storage is depleted, thereby decreasing the ATP formation resulting in the death of the parasite. Recurrence rate after surgical treatment ranges from 1% to 20% but is generally 5% or less in experienced centres. Preoperative treatment may decrease the risk of spillage and recurrence. During surgery, packing of the abdomen is important because rupture can result in anaphylaxis and seeding.

## CONCLUSION

To conclude, hydatid disease has varied clinical manifestations. High suspicion should prompt early radiological assessment as chest radiograph, ultrasonography and CT scan identify most cases. To summarize, a subcutaneous hydatid cyst should be considered as one of the differential diagnoses for soft tissue masses, particularly in patients who have lived in endemic areas. The treatment of choice remains surgical excision.

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