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## Unusual locations, presentations of hydatid cyst and their laparoscopic management

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### Abstract

**Background:** Hydatid disease (HD) is caused by the tapeworm *Echinococcus granulosus*. Hydatid cyst can involve all human parts and most common affected organs are liver and lungs. Incidence of unusual site is about 8-10%.

**Aim:** To study unusual locations, varied presentations of hydatid cyst in the human body and their laparoscopic management.

**Materials and Methods:** A retrospective study of HD was carried in a medical college between Jan 2015 and Dec 2019. A total 40 cases of HD were treated during this period. Information on clinical presentation and management were reviewed.

**Results:** Twenty three cases were of liver HD and seven were with hydatid lung disease. Ten cases of HD of uncommon locations and presentations were encountered during this period. Most patients were managed laparoscopically or assisted techniques.

**Conclusion:** We conclude that *Echinococcus granulosus* can affect any organ in the body from head to toe, and a high suspicion of this disease is justified in endemic regions. Can be managed laparoscopically successfully.

**Keywords:** Hydatid disease, echinococcosis, pancreas, laparoscopic, spleen, unusual locations

### 1. Introduction

Hydatid disease (HD) is a zoonosis which is endemic in the Middle East Africa, South America, New Zealand, Australia, Turkey, and Southern Europe, but foci are common in almost every part of the world including India where the highest prevalence is reported in Andhra Pradesh, Tamil Nadu, and Jammu and Kashmir. Hydatidosis is caused by the larval form of *Taenia Echinococcus*, which lives in the gut of dog, wild canines and carnivorous animals that represents the definitive host. It can involve also both domestic and wild animals. In this way Humans become the accidentally intermediate hosts by ingesting *Taenia* eggs. <sup>[1, 2]</sup> Then the slowly growing hydatid cysts can achieve a volume of several liters and contain many thousands of protoscolices. With time daughter cysts can be formed. <sup>[3]</sup> Infestation by *Echinococcus granulosus* in humans most commonly occurs in the liver (55-70%) followed by the lung (18-35%); the two organs can be affected simultaneously in about 5-13% of cases. Incidence of unusual sites is about 8-10%. Incidence of HD involving the spleen, kidney, peritoneal cavity, skin and muscles is about 2% each and incidence of the heart, brain, vertebral column, ovaries, pancreas, gallbladder, thyroid gland, breast, and bones involvement is about 1% each. <sup>[4]</sup> The clinical picture depends upon the involved organs, its effects on adjacent structures, complications due to secondary infection, rupture and anaphylaxis caused by hydatid cyst. This benign disease can cause substantial morbidity and mortality. The aim of this study is to highlight the fact that this disease should be suspected in cystic lesions involving any organ in the body, especially in endemic areas like India. Even though, hydatid cysts can affect any organ, the disease is uncommon in the organs cited here. To our knowledge, there are some case reports in the literature describing unusual locations of hydatid disease, but only few case series of unusual locations of echinococcosis have been published to date.

Herein, we present a retrospective study on this topic performed on 40 patients surgically treated for hydatid disease between 1995 and 2019 at our Department. A series of 40 patients with 10 unusually located hydatid cysts is presented and analyzed.

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## 2. Materials and Methods

A retrospective study was carried out in our medical college during Jan 2015 to Dec 2019. A total 40 cases of HD were treated during this period. Information of clinical presentation and management was reviewed. As a retrospective study, approval to use the data was obtained from the relevant authorities, but no formal ethical clearance was sought. The age and gender of the patients, the location of the echinococcal cysts, the type of surgical procedure, the length of postoperative stay, the cysts diameter, complications, the morbidity and mortality were examined.

Clinical features, examination, blood counts (eosinophilia), serological test (ELISA test), ultrasound, and computed tomography (CT) confirmed the diagnosis. All patients were given Albendazole in the pre-operative period and then operated

to minimize the chances of post-operative recurrence. Pain was most important presenting symptom. In two cases, presenting symptoms were due to compression on adjacent organs.

## 3. Results

Twenty three cases were of liver HD and 7 were of hydatid lung disease. Seventeen cases were with right lobe involvement and three were with both lobe involvements. Out of seven lung hydatid only one case was with bilateral lung involvement. Ten cases of hydatid cysts at uncommon locations and presentations were recorded as shown in Table 1. Eosinophilia was present in all ten cases. ELISA test (serology) was positive in seven cases out of ten cases. Eight cases were of primary HD of uncommon site and in rest two cases there was liver HD along with pelvic hydatid.

**Table 1:** Location, age and sex, different form of presentations, serology, imaging investigations, management and complications of all the ten unusual cases

Location	Age	Clinical features	ELISA	Imaging	Management	Complications
Peritoneal hydatidosis	58/M	Huge abdominal distension	+ve	Multiple cyst occupying whole abdomen	Laparotomy drainage and evacuation of fluid and cysts>300	Pelvic collection
Lt. liver lobe+Pelvis	35/F	Abdominal pain	+ve	Lt. liver cyst (7x6cms) Two pelvic cyst (9x3cms and 2.5x3cms)	Lap Deroofing of pelvic cyst+ Lap Pericystectomy for liver hydatid	No
Multiple ruptured liver cyst	25/M	Abdominal pain, distension, fever and jaundice	+ve	Multiple liver cyst with CHD compression and ruptured Lt lobe cyst	CBD stenting+ Laparotomy lavage & Drainage	Peritonitis
Rt. lobe liver + pelvic cyst abutting Urinary bladder	32/F	Chronic abdominal pain	+ve	Rt. lobe liver seg V and VI and 2.6x3x3cms Antero lateral wall urinary bladder	Lap Deroofing of liver hydatid+ cystectomy of pelvic hydatid	No
Anterior abdominal wall (Extra peritoneal cyst)	60/F	Abdominal lump	+ve	14x11cms Extra peritoneal cyst hanging into abdomen freely from anterior abdominal wall	Lap assisted excision of complete cyst	No
Intra muscular Hydatid	33/F	Painless left scapular swelling	-ve	17x16cms left scapula soft cystic mass-intramuscular hydatid	Cyst excision	No
Pancreatic Hydatid cyst	24/F	Abdominal pain with lump	-ve	12x7x7cms Exophytic hydatid cyst of pancreatic tail	Lap assisted Cystectomy	No
Splenic hydatid	42/F	Left hypochondriac pain	+ve	15x11x14cm lower pole splenic hydatid cyst	Lap spleen preserving cyst Deroofing	No
Splenic hydatid	18/F	Abdominal pain	+ve	8x9cms well defined hydatid cyst upper pole spleen	Splenectomy	No
Retro peritoneal Hydatid	46/F	Abdominal lump with pain	-ve	well defined large retroperitoneal cystic lesion involving Lt. hypochondrium and left lumbar region 18x17cms, craniocaudal extension 26cms	Laparotomy, decompression and marsupialisation cyst	No

Open surgery was done in 10 cases, 4 cases were managed by medical therapy and 26 cases managed laparoscopic /laparoscopic assisted techniques. Deroofing/marsupialization was done in most of the cases except in cyst which was adherent with important structure such as ureter, blood vessels or nerves. Pericystectomy /complete cystectomy were done where cyst was lying freely in abdominal cavity/ hanging from liver lobe. The mean length of postoperative stay was 7 days (range 3–12 days). All patients were given Albendazole 15 mg/kg/day for 4 weeks. In most of the cases, there were no post-operative complications except in one case where there was post-operative pelvic collection which was drained percutaneous (ultrasound guided). There was no mortality and superficial wound infection was present in two cases only. All patients were followed upto 6 months only. There was one recurrence in Rt. lobe of liver after

6 months for which open pericystectomy was done.

## 4. Discussion

Hippocrates was first to illustrate a liver hydatid cyst and pioneered techniques of treatment [5]. More than 80-90% of hydatid cysts occur in the liver, lungs, or both. Hydatid cysts have been reported infrequently in the spleen, kidney, peritoneal cavity, skin and muscles and rarely involve the heart, brain, vertebral column, ovaries, pancreas, gallbladder, thyroid gland, breast, and bones. [4] Clinical presentation of HD depends upon the size, site and depth of the lesion. Eosinophilia is expected in all patients with parasitic infestations, this was seen in all our cases. HD is diagnosed mainly by history, examination and by radiological imaging. Ultrasonography (USG) is the first line of screening for abdominal hydatidosis and it is especially useful in

the detection of cystic membrane, septa, and hydatid sand. It also forms the mainstay in the follow-up period, for the detection of any complication or recurrence per se. [6] CT scan best demonstrates cyst wall calcification and cyst infection. CT scan imaging is also a modality of choice in peritoneal seedling. [7,8] Combination of USG and CT scan is most helpful imaging diagnostic tools. [9] On CT their appearance varies: They may show a “spoke wheel” pattern or a water lily sign. When cysts are healed or in an inactive state, they appear as multiple cystic lesions or with calcification in the peritoneum. [10]

Different serological tests are done for the diagnosis, screening, and post-operative follow-up for recurrence. These tests consist of the hydatid immunoelectrophoresis, enzyme-linked immunosorbent assay (ELISA), latex agglutination and IHA test. [11] The ELISA test has a sensitivity of 80-100% and specificity of 88-96% in hepatic cysts, 50-56% sensitivity in lung HD and 25-65% in HD of other organs. But these tests may be negative because the capsule isolates the parasite from the host's immune system. In our study, serology (ELISA) was positive in seven out of ten cases.

#### 4.1. Pathophysiology of spread of disease

Usually parasites spread via portal blood stream. Other routes of spread may be lymphatic invasion by the parasite, and retrograde migration from the vena cava to the subclavian vein. [11] HD can also involve any organ of abdomen due to hematogenous route or due to peritoneal fluid circulation phenomenon.

Peritoneal cavity has a normal circulation of peritoneal fluid due to various compartments in abdomen. A small amount of peritoneal fluid continuously circulates normally in abdomen. The movement of the diaphragm and peristalsis of bowel regulate the movement of fluid in this circulatory pathway. It mostly flows up in the right paracolic gutter, which is deeper and wider than the left. It is partially cleared by the subphrenic lymphatics. Fluid stays in these watershed regions in the peritoneal cavity: The ileocolic region, the root of the sigmoid mesentery, and the Pouch of Douglas. The spread of HD can be along the areas of peritoneal fluid circulation and may result in spontaneous intraperitoneal seeding. [10]

Incidence of HD involving the spleen is about 2-2.5%. It can occur primarily or in association with hepatic, pulmonary, or multi-organ hydatidosis. [12] In our study we encountered 2 cases of isolated splenic hydatid cyst (5%).

A primary HD of the retro peritoneum is a distinct clinical entity

that must be considered when caring for a patient with a retroperitoneal mass in endemic regions. Retroperitoneal involvement was always thought to be secondary to rupture or spillage during surgery of liver hydatids. A primary retroperitoneal hydatid cyst without other organ involvement was first reported by Lockhart and Sapienza, [13] in 1958 and since then only few cases have been reported. We have reported one case of primary retroperitoneal hydatid managed surgically.

Incidence of peritoneal involvement is about 12%. Clinical features include flank pain, abdominal mass and non-specific symptoms such as nausea and vomiting. It should be treated after the diagnosis is confirmed without any delay because of secondary spillages due to rupture and other possible complications. [14] Primary hydatidosis is an extremely rare entity accounting for just 2% of all intra-abdominal hydatid disease [15]. Our study has one case of massive disseminated hydatidosis with Liver and pelvis involvement.

Pelvic HD is unusual, and the reported incidence is HD is about 2.25%. [16] Due to its location in a fixed cavity, it presents with pressure effects on adjacent organs such as the urinary bladder (most common) or rectum. Urinary symptoms may be retention of urine, frequency of micturition or as obstructive uropathy and renal failure. [17] We also had a 2cases of pelvic hydatid along with liver hydatid in our study managed laparoscopically.

Location in muscular tissue accounts for 2-3% of all cases. We have operated one case of scapular muscle hydatid. Scapular hydatid is one of the rare sites to be found.

Pancreatic hydatid cysts (PHC) are rare entities with incidence ranging from 0.14% to 2%. [18] PHCs are usually solitary (90%–91%) and distributed unevenly throughout the head (50%–58%), body (24%–34%) and tail (16%–19%) [19]. We have one case of pancreatic tail hydatid in our study operated successfully.

Complications occur in 5% to 40% patients with hepatic hydatid cysts and include the formation of small cystobiliary fistulas, cyst rupture into the biliary tree, biliary compression, cyst infection, hydatid allergy and intraperitoneal rupture. [20] The reported frequency of liver hydatid cyst rupture into the peritoneal cavity ranges from 1% to 16%. [21]. Rupture may result from trauma or may occur spontaneously from increased pressure of the cystic fluid. The main risk factors predisposing to rupture include young age, cyst diameter >10 cm, and superficial cyst location. We have one case of ruptured liver hydatid presented with features of peritonitis and jaundice with CBD compression by pre-operative ERCP stenting followed by laparotomy and peritoneal lavage.

**Table 2:** Other rare unusual site

Study	duration	cases	Surgical management	serology	complication
J.Prosalidis <i>et al.</i>	1967-1994 27 years	Total-540 cases Unusual-49 cases	All open surgery	27 cases	2 cases
Georgios D. Lianos <i>et al.</i>	1980-2013 33 years	Total-233 cases Unusual-18 cases		NA	11%
Enver O <i>et al.</i>	1983-1999 15 years	Unusual-49 cases		32 cases	11 patients
Pinar Polat <i>et al.</i>	1996-2001 5 years	Total-368 cases Unusual-85 cases			
Arif Hussain <i>et al.</i>	2000-2014 15 years	Total-527 cases Unusual-38 cases			
Kasim Caglayan <i>et al.</i>	2004-2008 5 years	Total-98cases Unusual-10 cases	Open -6 cases Lap-0	85% positive	2 cases
Majid Mustaque <i>et al.</i>	2005-2009 5 years	Total- 244 cases Unusual-20 cases		14 patient	1 case
S.Sacher <i>et al.</i>	2007-2012 5 years	Total-79 cases Unusual-8 cases	All open surgery	7 cases	2 cases
Our Study	2015-2019 5 years	Total-40 cases Unusual-10 cases	Open- 5cases Lap-5 cases	7 cases	1 case

**Table 3:** Incidence in various study are listed

Name of organs	Incidence (%)	Reported by
Brain	2	Greenberg <i>et al.</i> 2001
Thyroid	1	McManus <i>et al.</i> 2003
Breast	1	McManus <i>et al.</i> 2003
Heart	1	McManus <i>et al.</i> 2003
Gall Bladder	<1	Raza <i>et al.</i> 2003, Mushtaque <i>et al.</i> 2011
Pancreas	0.5-0.8	Palanivelu <i>et al.</i> 2007, Moosavi <i>et al.</i> 2007
Uterus and Adnexia	0.5-1	Palanivelu <i>et al.</i> 2007, Arora <i>et al.</i> 2005
Seminal vesicle	0.1-3	Safioleas <i>et al.</i> 2006, Vasileios <i>et al.</i> 2002
Bones and spine	0.1-3	Drimousis <i>et al.</i> 2006
Muscle	2	Arora <i>et al.</i> 2011
Skin and subcutaneous tissue	1-2	Zulfikaroglu <i>et al.</i> 2005, Dirican <i>et al.</i> 2008

## 4.2. Management

Treatment of HD is mainly surgical (open as well as laparoscopic). Laparoscopic procedure involves: Aspiration, installation of scolicalid agent, deroofting, removal of all contents and converting the cyst into a big size non-dependent cavity. The advantages of laparoscopic surgery over open surgery are- less operative duration, less intraoperative complication, less pain, shorter hospital stay and better cosmesis. However, pre and post-operative 1 month courses of Albendazole should be considered in order to sterilize the cyst, to decrease the chance of anaphylaxis, to decrease the tension in the cyst wall (thus reducing the risk of spillage during surgery) and to reduce the recurrence rate post-operatively.<sup>[11]</sup> Intra-operatively, the use of gauge pieces soaked with hypertonic saline or 10% betadine solutions were used to surround the cyst before opening the cavities. These scolicalid kill the daughter cysts and therefore, prevent further spread or anaphylactic reaction.<sup>[11]</sup>

While surgery still remains as the standard for cystic echinococcosis treatment, there have been a number of studies that suggest that percutaneous drainage of cyst - puncture, aspiration, injection of hypertonic saline and absolute alcohol, reaspiration with chemotherapy is more effective than surgery in terms of disease recurrence, and morbidity and mortality.<sup>[22]</sup> In the case of alternative medical therapy using chemotherapy alone, Albendazole is used with an adult dosage of 400 mg orally, twice a day for 1-5 months and a pediatric dosage of 15 mg/kg/day (maximum of 800 mg) for 1-6 months.<sup>[23]</sup>

Even though, mortality directly due to echinococcosis is very low, it can produce a very disabling morbidity. A mortality rate between 0.29% and 0.6% has been reported.<sup>[24]</sup> The recurrence rate of this disease is still relatively high accounting for about 10%.<sup>[25]</sup> We had one recurrence of liver hydatid after 6 months, managed surgically.

The present study confirms that echinococcosis can be found in any organ of the body and that hydatid disease still constitutes nowadays a real and relevant health problem. Additional measures, such as the education of farmers and general public are essential in the control of this zoonotic infection in animals and humans<sup>[26]</sup>.

## 5. Conclusion

We conclude that *E. granulosus* can affect any organ in the body from head to toe except hair and nails. A high index of suspicion is required for pre-operative diagnosis of HD in unusual locations, and it should be considered in the differential diagnosis for any cystic mass found in patients from endemic areas. Moreover, medical treatment should precede and follow the surgical intervention.

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