ORIGINAL RESEARCH PAPER
Volume - 12 | Issue - 03 | March - 2023 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

VARIOUS PRESENTATION OF HYDATID CYST IN ADULT POPULATION: A CASE SERIES FROM TERTIARY CARE HOSPITAL IN CENTRAL INDIA.

General Surgery

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ABSTRACT

Cystic echinococcosis (CE) is a zoonosis caused by the larval stage of the dog tapeworm Echinococcus granulosus. Liver and lungs being the most commonly affected organs whereas splenic infection is rarer and its primary involvement occurs in less than 2% of abdominal Cystic echinococcosis. The clinical spectrum of hepatic hydatid cyst as well as splenic hydatid cyst may include abdominal lump with vague dull aching pain, postprandial fullness, retrosternal burn, nausea, vomiting with fever. Usually patient has history of dogs, cattle as well as goats as their pet animals. Sero-diagnosis plays a pivotal role in evaluation of abdominal lump which may later reveal itself as hepatic, splenic or hepato-splenic hydatid cyst. Here, we present different cases of hepatic and splenic hydatid cyst for which a combination of haematological as well as abdominal ultrasonography and computed tomography (CT) were used for preoperative diagnosis.

KEYWORDS

INTRODUCTION

Hydatid disease is a systemic zoonotic disease caused by tapeworms of genus *Echinococcus*. Infection in humans occurs accidentally by the ingestion of parasite eggs excreted in dog feces through hand-to-mouth transfer. Although splenic hydatid disease is a rare clinical entity with a prevalence of 2.5%–5.8%, the spleen is, after the liver and lungs, the third most common organ involved [1].

The life cycle of Echinococcus is indirect and involves two hosts, one definitive carnivore host and the other intermediate herbivore host [2] .The disease occurs when humans act as an accidental intermediate host and ingests viable oncosphere containing eggs, which have been shed in the faeces of the definitive host. The oncospheres invade the intestines, to enter the vasculature and develop into hydatid cysts in any organ or tissue, where a variety of symptoms can be produced. However, the liver acts as the first filter for hydatid larvae, making it the most commonly affected organ followed by lung.[3]

Humans are usually infected during childhood, and develop symptoms after several years from infection. Symptoms are related to enlargement or complication of the cyst(s) (i.e., rupture or infection).The organs most commonly involved are the liver (70%) followed by the lungs (25%), while other organs (e.g., brain, bones, spleen, kidneys) are more rarely involved, accounting for approximately 5% of cases.[4]

Depending on the stage of the disease, a hydatid cyst in the spleen can share very similar imaging findings with the splenic cysts of other etiology. Even with the use of modern radiological imaging techniques, this form of hydatid disease is a rare occurrence and may pose a diagnostic challenge. [5] Surgical management employs a wide range of interventions, from splenectomy to organ-sparing surgical procedures.

The radical surgical removal of the cystic lesion remains the mainstay of treatment with a high success rate [6]. Chemotherapy, with benzimidazole compounds has also been used with some success to sterilize the cyst, decrease the chance of anaphylaxis, and reduce the complications and recurrence rate post-operatively. In recent years, a third treatment option was introduced (PAIR, puncture, aspiration, injection, and re-aspiration) and is indicated for patients who cannot undergo surgery [7].

Case Presentation

The five patients described in this report were seen and evaluated in the Department of General Surgery in SAIMS Indore between September 1, 2021 to February28, 2022 for complaints of abdominal pain and gastrointestinal (GI) manifestations. Patients were interviewed and examined, after which blood samples were processed and radiological investigations were done

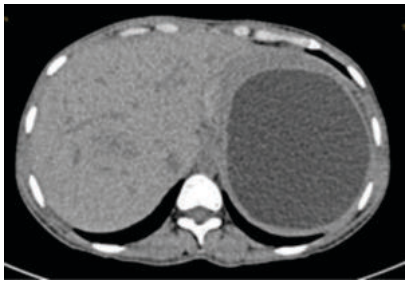
Pre-operative diagnosis of hepatic cystic lesions was made ultrasonically and confirmed by a computer tomography (CT) scan. After completion of this evaluation process surgical consultation was sought. All five patients were successfully managed surgically and all are asymptomatic till date.

Case 1:-

A 23 years old female, came with complains of pain in abdomen since 4-5 yrs with increase severity since 10-12 days. Pain was initially in left hypochondrium and radiated to whole abdomen , which was insidious in onset, gradual in progression and not associated with fever , cough, vomiting ,constipation with no other significant history.
On examination, it was noted that the per abdomen was soft, non tender, bowel sound present with palpable spleen upto 10 cm from below costal margin ,medially upto midline near umbilicus, hepatomegaly 5cm from below costal margin with guarding and rigidity being absent.

On further evaluation CECT ABDOMEN- two matted thick walled smoothly marginated hypodense cystic lesion were noted in right lobe of liver , measuring approx 11.6*9.6*10.3* cm around 570 cc and 9*7.7*6.7 cm around 230 cc. The lesion shows multiple curvilinear hyperdense thin membrane giving rise to **water lily sign**.

This lesion is compressing to the gall bladder .Post contrast , it does not show any internal enhancement-may suggest hydatid cyst **(type 2)**. Similar morphological cystic lesion having peripherally daughter cyst is noted in the upper pole of splenic parenchyma, which measures approx 14*14* 11.6 cm around 1140 cc.

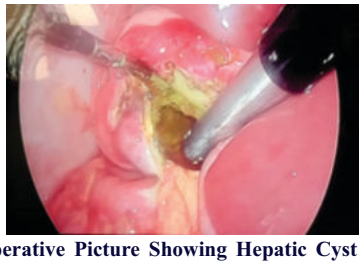


(1) Splenic Hydatid Cyst

(2) Hepato-Splenic Hydatid Cyst

(3) Coronal Section Showing Hepato-splenic Hydatid Cyst

We performed laparoscopic marsupilization of splenic cyst and hepatic cyst with partial pericystectomy with omentopexy of splenic cyst with abdominal drain placed in hepatic cyst cavity under general anaesthesia.



(4) Intra-operative Picture Showing Hepatic Cyst With Cysto-biliary Communication

Patient had drain output

POD	QUANTITY (in ml)	TYPE
POD 0	280	Bilious
POD 1	250	Bilious
POD 2	180	Bilious
POD 3	140	Bilious
POD 4	100	Bilious
POD 5	80	Bilious

Patient was discharged with drain in situ.

MRCP was done on post operative day 15 (outpatient basis) which revealed relative reduction in hepatic hydatid cyst of about 90% and approximately 55-60% reduction in splenic hydatid cyst.

Case 2:-

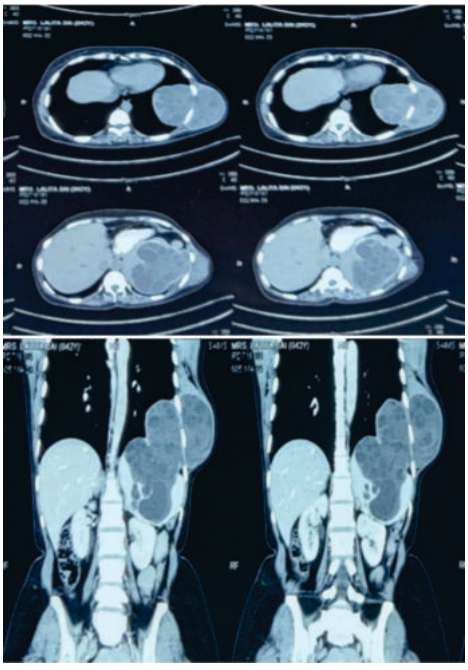
A 42 year female presented with Swelling in lateral aspect of left chest wall since 8 months. Swelling was over lateral aspect of left chest wall which was insidious in onset, gradually increasing in size of around 10x8cm with no pain.

Patient had history of exploratory laparotomy with marsupilization and omentoplasty done for Left Lobe liver hydatid cyst in 2011. On post operative day 3, she developed intestinal obstruction for which Re-exploration with adhesiolysis was done.

On examination, there was bucket handle scar mark in subcostal region and midline incision scar mark with no palpable organomegaly. A swelling of around 10*6 cm over lateral aspect of left chest wall from 5^{th} to 7^{th} intercostal space and extending from anterior axillary line to posterior axillary line. On palpation swelling had no local rise of temperature and tenderness, soft in consistency, non mobile, fluctuation test positive, transillumination test negative.

On ultrasonography it was revealed that multiple well defined anechoic clustered cystic lesions were noted involving most of the splenic parenchyma as well as an extension into the left hypochondrium and left lumbar region with few of them show peripheral calcification suggestive of splenic hydatidosis leading to peritoneal hydatidosis.

On CECT abdomen it was noted that there is lobulated cystic lesion involving spleen, diaphragm and extending in left lower chest cavity and across the chest wall into the subcutaneous planes. The lesion shows multiple variable sized daughter cyst suggestive of diaphragmatic /transdiaphragmatic spread of splenic hydatid disease with extrathoracic extension along the chest wall **(TYPE3/CE2)**



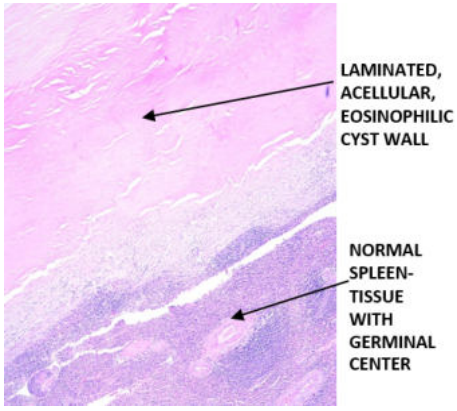
(5) CECT showing transdiaphragmatic spread of splenic hydatid disease with extrathoracic extension along the chest wall .

Patient was taken for Exploratory Laparotomy with omental adhesiolysis with Splenectomy done along with excision of hydatid cyst with Repair of diaphragmatic defect.

(6) Spleen and Hydatid cyst identified with Hydatid cyst

(7) Drainage of daughter cyst

(8) Specimen showing resected part of omentum, daughter cyst and part of spleen.



(9) 4x Magnification

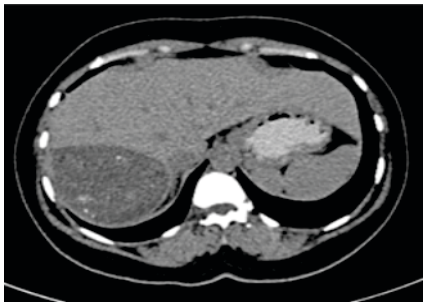
Post operative period was uneventful. Patient had mild serous fluid discharge from lateral chest wall wound which was reduced gradually. Patient discharged on stable condition on Tablet ALBENDAZOLE 400mg twice a day to be continued for 3 months.

(10) Post-operative pictures showing abdomen and chest wall closure with corrugated drain kept in situ.

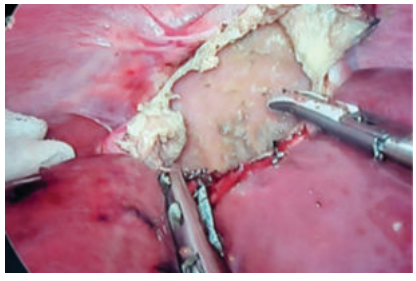
Case 3:-

A 35 years old female, was admitted to our hospital with complaint of pain in epigastrium since 1 year. Pain was initially at epigastric region and radiated to left hypochondrium, which was insidious in onset, gradual in progression and associated with post prandial fullness , vomiting and retro-sternal burn.

Patient had no comorbidities with no history of addictions. On examination, per abdomen was soft, non tender, bowel sound present with no palpable organomegaly. Echinococcus IgG antibody was done which resulted out to be negative. The patient was planned for CECT abdomen which revealed a low attenuation lesion is seen in the right hepatic lobe involving the segment VI/VII measuring approximately 7.2 x 8.6 x 6.9 cm in AP, TR and CC axis respectively abutting the inferior vena cava and right branch of the main portal vein.**(type 1/ CE1)** Multiple small internal foci of calcifications are noted within this region. Multiple small fat density foci are also noted within this lesion.



(11) Right lobe hepatic hydatid cyst



(12) Intraoperative picture showing marsupilization of cyst

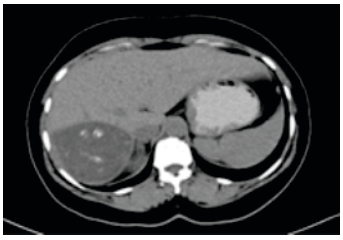
(13) Picture showing hydatid cyst fluid

Laparoscopic hydatid cyst marsupilization was done. Post operatively patient was stable.

Case 4:-

A 26 years old female came with complains of pain in right hypochondrium since 2 months. Pain was initially at right hypochondium region and radiated to whole abdomen, was insidious in onset, gradual in progression and associated with on/off fever since 1 month.

On examination, it was noted that the per abdomen was soft, non tender, bowel sound present with no palpable organomegaly. Cect abdomen revealed a low attenuation lesion seen in right hepatic lobe approx. 9.2*9.1*6.9 cm with thick wall and internal floating membrane with dense focal of calcification also noted within lesion . The lesion is reaching upto sub capsular space - suggestive of large hepatic cyst in right hepatic lobe.**(TYPE2/ Ce3)**

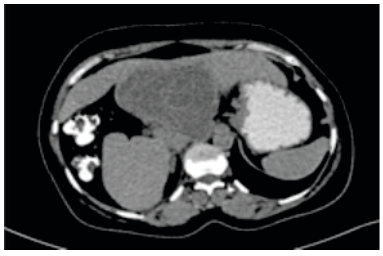


(14) Cect showing liver hydatid cyst

Laparoscopic hydatid cyst marsupilization was done under GA. Post operatively patient was stable.

Case 5:-

A 55 year old female presented to us with complain of pain over right hypochondrium since 2 years which increased progressively since 1 year. Pain was insidious in onset, gradually progressive. On examination, it was noted that the per abdomen was soft, non tender, bowel sound present with liver palpable upto 12cm below the costal margin .Cect was suggestive of liver being enlarged measuring 17 cm. There is evidence of a well circumscribed cystic lesion with floating internal membranes and no internal vascularity noted in left lobe of liver segment (II and III) suggestive of hepatic hydatid. There is evidence of another hyperechoic lesion in segment VII of liver measuring 4.3 x 3 cm suggestive of old calcified hydatid lesion. There is evidence of a well defined cystic lesion with wall calcification is seen in segment VII and VIII of right lobe of liver and extending/ protruding in right sub-diaphragmatic region**.(type 5/CE 5)** It measures approx. 8x6.8 cm. Another cystic lesion with internal septations/membranes noted along inferior aspect of segment IV b of left lobe of liver- protruding in subhepatic region **(type 2/CE3)**. It measure approx. 7.9x9.6 cm. Similar morphology tiny cystic lesion of size 1.2x1.0 cm is seen in the body/tail region of the pancreas.



(15) Cect showing bilateral hepatic hydatid cyst

(16) Coronal section showing bilateral hepatic hydatid cyst with lesion in pancreas

(17) Intraoperative picture showing hepatic hydatid cyst

Patient was taken for exploratory laparatomy with hydatid cyst excision with omentopexy.

DISCUSSION:-

Hydatid disease is a systemic zoonotic disease caused by tapeworms of the genus *Echinococcus*. Usually it is a disease of sheeps, dogs as well as cattles but accidentally may infect humans by the ingestion of parasite eggs excreted in dog feces through hand-to-mouth transfer. Most common organ involved is liver followed by lungs and spleen being the third most commonly involved organ with prevalence rate of 0.5 to 2% followed up by kidneys and brain. [8]

Rarely, primary pancreatic hydatid cyst is observed with an incidence between 0.14% to 0.2%. Pancreatic involvement occurs mainly because of hematogenous dissemination, although lymphatic or direct passage from the intestine also plays a key role.

The **clinical spectrum** of hydatid cyst depends from the organ involved. Although there is huge percentage of asymptomatic patients, which gets confirms once the patient gets hematological as well as radiological investigation making it a accidental diagnosis of hepatic hydatid cyst. The most frequent symptom is pain located at the level of the right hypocondrium associated with various non specific symptoms like vomiting, back ache, postprandial fullness, heartburn, constipation etc. Apart from this other symptoms depend on system or organ involved like hydatid cyst of the hip extending to the gluteal region along with ischiorectal fossa may present as perianal mass, [6]. Cardiac hydatid disease accounts for 0.5–2% of all cases of hydatid disease may remain asymptomatic for many years or show symptoms depending on the location and size of the cysts, such as chest pain, dyspnoea, arrhythmia and anaphylactic reaction. **[7]**

Ultrasonography and computed tomography scan often lead to diagnosis in more than 90% of cases, and can pinpoint the etiology. The sensitivity of tri-phasic enhanced multi-slice CT is superior to that of sonography. The imaging appearance has been evolved over the time and is divided into two phases: pre-suppurative and suppurative. In the pre-suppurative phase, images are heterogenous, hypodense, with irregular contours, poorly demarginated, and may simulate tumor, especially when cyst are multiple and small.

During the suppurative phase, images are hypo- or anechogenic, sometimes multiloculated, with rounded contours, that are clearly delineatedby a more or less thick capsule. During this phase, sonographic images may have a typical "target" appearance. After contrast injection, peripheral enhancement forms a hyperdense border, the so-called "ring sign" without central enhancement.[9]

The presence of bile plays a essential diagnostic role and helps to define the communication with the biliary tract, either by contiguity, or because of ischemia/necrosis of the bile ducts. It may be classified as either **communicating or non-communicating** with the biliary tree, according to the presence or absence of bile in the drainage. When a cyst contains bilious secretion, biliary obstruction should be suspected, like choledocholithiasis. Biliary MRI or cholangiography should be used to visualize entire biliary tree to assess associated disease[8] .

Serology tests such as ELISA or immunoblotting is used which is 80-100% sensitive for liver cysts and 50-56% for lungs and other organs. False positive reactions may occur in persons with other tapeworm infections, cancer, or chronic immune disorders. Patients having calcified or dead cysts are usually sero-negative.

Detection of IgG antibodies implies exposure to the parasite, while in active infection high titers of specific IgM and IgA antibodies are observed.

The counter-immunoelectrophoresis has the highest specificity (100%) and high sensitivity (80 – 90%). CASONI TEST has been used most frequently in the past but is abondoned because of low sensitivity. WHO introduced a standardized classification of Ultrasonography.

Surgery is avoided in cases of multiple cysts localised in several organs and if surgical facilities are inadequate. The chemotherapy and **PAIR technique** (puncture-aspiration-injection-respiration) is a alternative treatment, especially in inoperable patients and for cases with a high surgical risk. Cysts with homogeneously calcified cyst walls does not need surgery and is managed conservatively.

The surgical procedures can be divided into two large groups, a conservative group and a radical one.

Conservative procedures are safe and technically simple, and are useful in the management of uncomplicated hydatid cysts. Marsupialization is the most common used procedure.

Radical surgical procedures include cystectomy, pericystectomy, lobectomy and hepatectomy. Radical procedures have lower rate of complications and hence used commonly.

Laparoscopic offers a lower morbidity outcome and a shorter hospital stay and it is also associated with a faster surgery with better visual control of the cyst cavity under magnification which allows a better detection of biliary fistula. Disadvantage of laparoscopy is the lack of precautionary measures to prevent spillage under the high intraabdominal pressures caused by pneumoperitoneum, allergic reactions are more common in laparoscopic interventions due to peritoneal spillage.[10]

Medical treatment of hydatid liver cysts are benzoimidazole, carbamates, such as mebendazole and albendazole. These drugs decrease the size of cyst. The factors for success seem to be the ability of the drug to penetrate the cyst wall and the persistence of adequate levels of the active metabolites. Albendazole seems to be more effective owing to better penetration and absorption. Side effects of Albendazole therapy are: mild abdominal pain, nausea, vomiting, pruritis, dizziness, alopecia, rash and headaches. Occasionally, leucopoenia, eosinophilia, icterus, and mild elevation in transaminase levels are seen.

CONCLUSION:-

Hydatid disease is a health problem in endemic countries. Usually patients have a history of being associated with dogs, sheeps and cattles. The liver is the most common site for hydatid disease (75%), lungs (15%), the spleen (5%),and other organs (5%) like intracardiac, foot, eyes etc. **Here in our study**, we conclude that there can be different clinical presentation depending upon the organ involved, commonly abdominal lump with right or left hypochondric pain , post prandil fullness, nausea, vomiting, fever etc. Intracardiac hydatid cyst results into chest pain, dyspnoea, arrhythmia and anaphylactic reaction. Various uncommon presentation such as shortness of breath, cough etc is also seen in pulmonary echinococcosis. Ultrasonography and computated tomography associated with serodiagnosis remains the mainstay in investigation. Often surgical management remains mainstay line of treatment, but technique like PAIR and medical management are used in cases of inoperability.

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