

## Hydatid Cysts Presenting in Uncommon Sites – An uncommon case series

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### **Abstract**

Hydatid cysts are commonly seen in the lung and liver. In the last 3 years in our institute we had a total of 25 patients of Hydatid cysts which included 13 cases of liver, 3 lung, 2 intramuscular, 1 submandibular gland, 1 recurrent intraperitoneal hydatid cyst (with history of previous liver hydatid surgery 6 years prior) and one case each of hydatid cysts at six uncommon intra abdominal sites- pancreas, kidney, ovary, mesentery, spleen and appendix.

**Key words:** Hydatid cysts, intra-abdominal, Intra-peritoneal, uncommon sites

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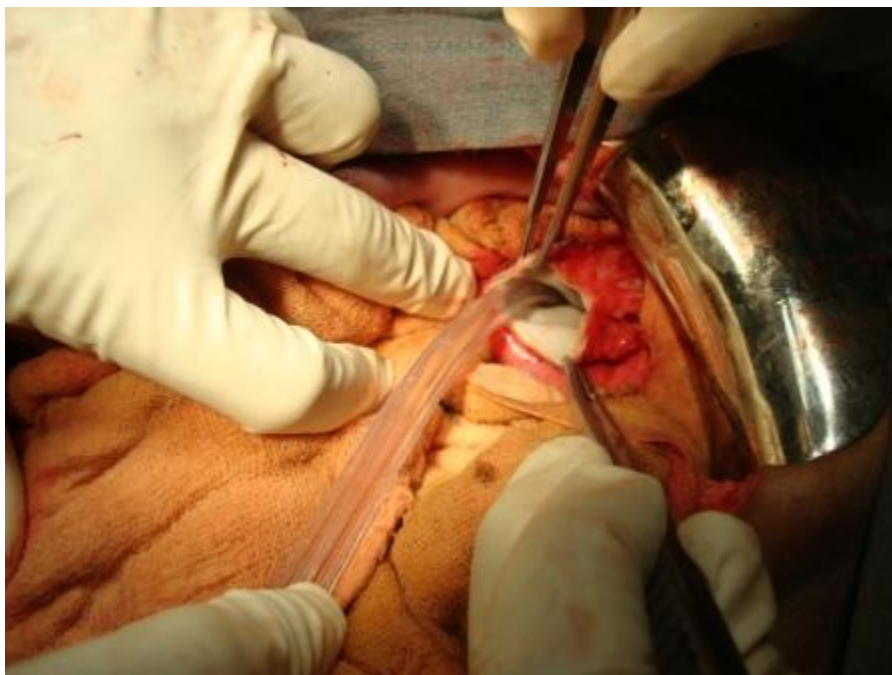
The cases are described of hydatid cysts presenting at uncommon sites:

### **Case presentations:**

**Case 1-** A twenty six years old male patient was admitted with pain in the left hypochondrium for one month. The pain was non-radiating with no aggravating or relieving factors. The liver was palpable 2 cm below the costal margin and was firm and non tender. The rest of the general and systemic examination was normal. USG abdomen showed a large cyst measuring more than 8 cms in size, close to the pancreatic tail and a small isoechoic calcified cyst in the right lobe of liver. CT abdomen showed two calcified Hydatid

cysts of sizes 3.3 x 3.1 cms and 3.1 x 2.9 cms in the liver and a well defined lesion in the region of body and tail region of pancreas of size 8.9 cm by 6.6 cm suggesting the possibility of hydatid cyst in Pancreas [post contrast] with a differential diagnosis of Pseudo cyst or cystic neoplasm of pancreas. The patient was given pneumococcal vaccine and H. influenza vaccine pre-operatively and taken for explorative laparotomy. Intra operatively two cysts in the liver and one large cyst in pancreas were seen.

**Figure 1: The liver cysts and pancreatic cyst were excised. Roux en Y anastomosis was done. Histopathology of the pancreatic and liver cysts confirmed the diagnosis of hydatid cyst.**



**Figure 1: Intraoperative photograph of hydatid cyst of pancreas.**

**Case 2-** An eight year old boy presented with abdominal pain and low grade fever for 3 days. He had similar complaints six months earlier. On examination he had a soft abdomen. On investigation he had normal hematological parameters, normal urine examination, liver and kidney function tests and a normal chest x ray. USG abdomen done outside revealed a cyst in the right kidney measuring 5.1 x 4.3 cms and occupying the middle and lower pole. CT

abdomen showed a well defined, none enhancing, multiloculated lesion, and measuring 4.7 x 5.1 x 3.8cms, occupying the middle and lower pole of the right kidney and displacing the adjacent calyces without any involvement. No calcification was seen. The liver was normal. The radiological diagnosis was right multilocular cystic nephroma/ complex cortical cyst. The patient was taken for surgery. A right nephroureterectomy was done and the

specimen was sent for histopathology which grossly showed a hydatid cyst of the kidney,

[Figure 2] which was also confirmed on histopathology.



**Figure 2: Gross photograph showing hydatid cyst in the kidney**

**Case 3-** An eighteen years old female presented with a gradually increasing abdominal lump, associated with pain on and off since 10 years. On examination the abdomen appeared distended and a large, mobile, non tender, cystic lump was palpable measuring about 14 x 9 cm in the right hypochondrium and epigastric region and extending to the right iliac region. CT scan abdomen showed a large well defined, thick walled multi cystic lesion of size 8.7 x

14.8 x 13.8 cm in the umbilical and epigastric region, partially extending in the right hypochondriac and lumbar region. Multiple small cysts seen were seen within the lesion. There was no calcification noted within the lesion. The liver was normal. A radiological diagnosis of mesenteric cyst or ? hydatid cyst was given. The patient was taken for exploratory laparotomy. Intraoperatively a cystic trans illuminable lump was seen in the abdominal cavity,

adherent to the mesentery and inferior surface of the liver. The cyst was excised intact and sent for histopathology. There were no other intra abdominal cysts.

Grossly the cyst showed prominent blood vessels on its surface and its cut surface was multiloculated and showed the typical egg white inner lining of hydatid cyst.

**Figure 3: Histopathology confirmed hydatid cyst of the mesentery**



**Figure 3: Gross photograph of mesenteric hydatid cyst.**

**Case 4** - A thirty five yrs old female presented to the gynecology department with history of menorrhagia for 1 year and spasmodic abdominal pain for 6 months. On abdominal examination a cystic mass

25x10cm was felt in the suprapubic region extending upto the umbilicus. The lower border of the cyst could not be reached. Per vaginal examination revealed a bulky uterus and a large cystic left ovary. The right ovary

was also enlarged. A clinical diagnosis of Fibroid uterus with left ovarian cyst was made. Ultrasonography of the pelvis showed a posterior wall fibroid with a large? Ovarian cyst. CT abdomen showed a hydatid cyst in the right lobe of liver, along with a large ovarian cyst measuring 16x11x15cms and a posterior wall fibroid. Intra operatively a large hydatid cyst measuring 20 x 10 cms was seen in the

pelvic region along with a small intra peritoneal cyst and 2 cysts in the liver. The contents of the liver hydatid cysts were aspirated and removed, and the cavity was irrigated. Total abdominal hysterectomy with bilateral salpingo-oophrectomy and removal of the small intra peritoneal cyst was done. Histopathology confirmed bilateral ovarian hydatid cysts with a fibroid uterus. (Figure 4)



**Figure 4: Gross photograph showing bilateral hydatid cysts in the ovaries**

**Case 5-** A thirty years old male presented with abdominal pain and weakness for two months, cough for 15 days, fever for 8 days and vomiting for 3 days. He gave a history

of pet dog contact 12 years prior. On examination he had a soft, non tender abdomen. His hemoglobin was 8.9g%, TLC 15,400/cumm, and DLC showed 84%

neutrophils. Liver function tests were normal. CT scan showed a large cyst in the liver, multiple cysts in the spleen and in the peritoneum. The patient was taken for laparotomy where multiple cysts were seen in the liver, spleen, perisplenic area, pelvis, intraperitoneally and adhering to the appendix wall. Splenectomy and appendicectomy were done. Intra pelvic and intraperitoneal cysts excision was done. The

contents of the liver hydatid cysts were aspirated and daughter cysts removed, and the cavity was irrigated. The specimens were sent for histopathology. The spleen showed multiple hydatid cysts the largest measuring 3.5 x 2.5 cm. The appendix showed a thick walled cyst in its wall and in the mesoappendix measuring 2.5 x 2 cms. Histopathology confirmed the splenic and appendiceal hydatid cysts. (Figure 5)



**Figure 5: Gross photograph showing multiple hydatid cysts in the spleen. Inset in lower left corner shows hydatid cyst in the appendix of the same patient.**

### **Discussion**

Hydatid cysts are caused by the cestode parasite *Echinococcus granulosus*. The life-cycle of *Echinococcus*

*granulosus* occurs between domestic or wild carnivores, such as dogs, foxes, wolves, jackals, hyenas and cats (definitive hosts), and sheep, goats, cattle, pigs, yaks or other

farm animals (intermediate hosts). Cystic echinococcosis is principally maintained in a dog–sheep–dog cycle. Carnivores become infected when they ingest the organs of intermediate hosts that harbour the larval stages of the parasite (hydatids, or hydatid cysts). In the carnivore, the cysts develop into adult worms and live in the intestines where they produce eggs that are passed in the faeces, contaminating the ground. Intermediate hosts ingest eggs in the contaminated ground, the eggs develop into cysts, and the cycle continues. Humans are accidental intermediate hosts and become infected through the ingestion of soil, water or food (e.g. vegetables) contaminated with the parasite's eggs shed in the faeces of the carnivores. Humans can also be infected by hand-to-mouth transfer of eggs after contact with the contaminated fur of a carnivore (most commonly, a dog).<sup>1</sup>

The eggs hatch in the human duodenum and commonly invade the liver (55-70%) and lungs (18-35%) through the blood stream. It can also affect the brain, heart, kidney, ureter, spleen, uterus, fallopian tube, mesentery, pancreas,

diaphragm and muscles.<sup>2</sup> The larvae lodge within the capillaries and first incite an inflammatory response composed principally of mononuclear leukocytes and eosinophils. Many such larvae are destroyed but others further develop into hydatid cysts. The cysts grow at the rate of 1 to 2 cms per year.

Isolated pancreatic hydatid cyst is rare and is often associated with hepatic involvement as seen in our case. Less than 1% of hydatid cysts are found in the pancreas.<sup>3</sup> They have different locations - Head (57%), Body (24%) and Tail (19%). Occasionally they may be revealed by a complication such as rupture into biliary tree, peritoneal cavity or gastrointestinal tract, abscess formation, and compression of splenic vein causing portal hypertension. Cysts may also lead to acute pancreatitis by obstructing the pancreatic duct.<sup>4</sup> The diagnosis of pancreatic cystic lesion is by Ultrasonography, CT scan or MRI. The aim of surgery in pancreatic disease is to maintain the pancreatic exocrine and endocrine functions. The modalities of surgical treatment used are partial or total

cystectomy, pericystectomy, cystoenteric anastomosis, marsupialization, external drainage and distal pancreatectomy for hydatid cyst in the tail.<sup>5</sup>

Kidney involvement in echinococcosis is extremely rare, accounting for only 2-3% of cases.<sup>6,7</sup> It occurs in adults with a median age of  $46.5 \pm 13.7$  years and is extremely rare in children, with only isolated case reports of renal hydatid cyst occurring in children.<sup>8,9,10</sup> It remains clinically silent for a long time and only presents at the stage of complications. The diagnosis is essentially radiological. Renal hydatid cyst raises therapeutic problems due to its complications, which sometimes make kidney-preserving cyst resection surgery difficult. The clinical features in adults may include pain (80%), flank mass (42%), hydaturia (22%), haematuria (13%), urinary tract infection (6%), and hypertension (3%).<sup>10</sup> Children may present with pain or abdominal mass.<sup>8</sup> They may also have associated hydatid cysts in the liver or lungs.<sup>9</sup> Eosinophilia and positive hydatid serological tests may be helpful in making a clinical diagnosis. Treatment of renal

hydatid cysts is essentially surgical.<sup>7</sup> Our case was an 8 year old boy whose CT scan abdomen reported a non calcified right kidney cyst suggestive of multilocular cystic nephroma, hence it was not diagnosed clinically, and only picked up after surgery on histopathology.

Isolated mesenteric hydatid cyst is very rare, as was seen in our case. It is usually secondary to spontaneous or iatrogenic rupture of liver or splenic cyst.<sup>11,12</sup> The patients usually present with chronic abdominal pain, unless there are complications in the cyst. The common complications are hydatid peritonitis (due to rupture of the cyst responsible for anaphylactic reaction), infection of the cyst and compression of adjacent organs responsible for an occlusive syndrome.<sup>11</sup> The treatment of choice is principally a careful and complete surgical excision. The diagnosis of mesenteric hydatid cyst should be considered in patients with a cystic mass, who live or have lived in a geographic region that has a high risk for *Echinococcus granulosus*, or visited an endemic area.<sup>13,14</sup>



The incidence of primary hydatid cyst in the pelvic region is very rare and ranges from 0.2 -0.25%.<sup>15</sup> When it presents in the ovary it may mimic an ovarian neoplasm. When the hydatid embryo escapes the pulmonary circulation and enters the systemic circulation, it can enter the female reproductive system.<sup>16</sup> The common clinical problems of hydatid cysts in gynecology are menorrhagia, obstructed labor, lump in abdomen, sterility and retention of urine. Pelvic echinococcosis in women remains difficult to diagnose with sonography. CT scan is superior to detect extra hepatic disease.<sup>17</sup> This fact was endorsed in all our cases described above.

Secondary splenic hydatid disease usually follows systemic disseminated or intraperitoneal spread following a ruptured hepatic hydatid cyst.<sup>18, 19</sup> Splenic hydatid cysts occur in 1.5% to 3.5% of all cases of abdominal hydatidosis.<sup>19</sup> They are usually asymptomatic but may present as a painful mass in the left upper abdomen. Its complications are mainly due to compression, infection, intra-abdominal rupture, anaphylaxis, and secondary

hydatidosis, fistulization to colon, and rupture into the thorax. The main differential diagnoses of splenic hydatidosis are any splenic cystic lesions such as pseudocyst, abscess, hematoma, and cystic neoplasm. Splenectomy has been the traditional treatment of choice for splenic hydatid cyst.<sup>20</sup> Our patient also had an associated hydatid cyst in the appendix. This is because intraperitoneal hydatid cysts usually develop secondary to spontaneous or iatrogenic rupture of hepatic, splenic, or mesenteric cysts. Dissemination occurs either by lymphatic, or systemic circulation.<sup>21</sup>

None of our cases described above had any previous history of hydatid cyst. The patient with liver, spleen and appendix hydatid cyst had a past history of a handling pet dog. None of our patients had peripheral eosinophilia. Three of our cases of pancreatic, ovarian and spleen-appendix hydatid cysts had coexisting liver hydatid cysts also. All our cases were treated by surgical excision, followed by oral albendazole treatment. They were also counseled on the importance of personal

hygiene and simple measures like hand washing, to prevent such diseases. The patients are doing well and have not reported back with recurrence at the same or other site so far.

**Conclusion:** Hydatid cysts can present at unusual sites and cause significant morbidity, and hence should be kept in mind as a differential diagnosis in endemic areas. Patients and their relatives should be educated on basic hygiene and measures like hand washing to prevent this disease.

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