

# Unusual Presentation of Hydatid Cyst

Shiffali Sarngal<sup>1</sup>, Shivani Gandhi<sup>2</sup>, Swati Arora<sup>3</sup>, Shaveta Sharma<sup>4</sup>

## ABSTRACT

Hydatid cyst is a zoonotic disease that most commonly occurs in liver and lungs. Here, we present five cases of hydatid cyst occurring in axillary subcutaneous region, adnexal region, ovary, gallbladder, and pancreas. *Echinococcus* should be considered in the differential diagnosis of any cystic lesions in any anatomic location, with or without viscera involvement particularly in endemic areas.

**Keywords:** Disease, *Echinococcus*, Hydatid.

*Euroasian Journal of Hepato-Gastroenterology* (2022): 10.5005/jp-journals-10018-1369

## INTRODUCTION

Hydatid cyst is a zoonotic disease caused by larval stage of *Echinococcus*. Man is an accidental host and does not play role in biological cycle of the worm. It frequently involves the liver and lungs. Although rarely known to occur in other body areas like muscles, brain, bone, and breast, it is extremely rare in female reproductive system and subcutaneous tissue. There are different modes of presentation of disease. Involvement of organs can be primary or secondary following liver or lung involvement.

In this article, we present a series of five cases of hydatid cyst disease found in axillary subcutaneous region, adnexal region, ovary, gallbladder, and pancreas.

## MATERIALS AND METHODS

We report five cases of hydatid disease (Table 1) presenting with cysts at unusual locations, namely, axillary subcutaneous region, adnexal region, ovary, gallbladder, and pancreas.

## RESULTS

### Case Studies

#### Case 1

A 52-year-old male with a slowly growing painless axillary mass lesion was presented to department of surgery. On physical examination, he was afebrile and had a painless, smooth, soft-to-cystic mobile mass of 3 cm in diameter in the left axilla. There was no other local sign of inflammation or pus discharge from the lesion. Breast and systemic examination was unremarkable. Patient was advised routine blood investigations and FNAC of the mass lesion. Marked eosinophilia (eosinophil count 12%) was detected in the peripheral blood smear. FNAC was done using

**Table 1:** Depicting the location of hydatid cyst

Sl. No.	Location	Age/sex
1	S/C axillary region	52 years/M
2	Adnexal region	43 years/F
3	Ovary	34 years/F
4	Gallbladder	42 years/F
5	Pancreas	45 years/M

<sup>1-4</sup>Department of Pathology, GMC Jammu, Jammu and Kashmir, India

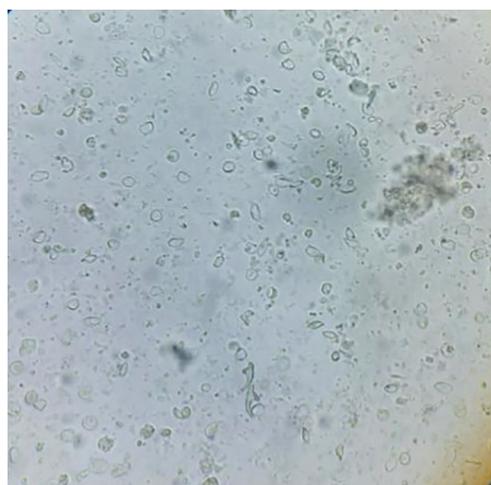
**Corresponding Author:** Shaveta Sharma, Department of Pathology, GMC Jammu, Jammu and Kashmir, India, e-mail: drshavetasharma@gmail.com

**How to cite this article:** Sarngal S, Gandhi S, Arora S, *et al.* Unusual Presentation of Hydatid Cyst. *Euroasian J Hepato-Gastroenterol* 2022;12(1):31–34.

**Source of support:** Nil

**Conflict of interest:** None

25-gauge needle yielded about 10 mL of clear, watery fluid and swelling decreased in size. Smears prepared from the centrifuged deposits of fluid and were then stained with MGG (May–Grunwald–Giemsa) and Pap (Papanicolaou) stain. Microscopic examination of the smears demonstrated many scolices with attached hooklets along with scattered hooklets and inflammatory cell infiltrate in the background. Fragments of acellular material with parallel striations which represent the remnants of laminated membrane were also seen. The lesion was later confirmed on histopathological examination (Fig. 1).



**Fig. 1:** Microphotograph of smears showing a high power view of hooklets

**Case 2**

A 43-year-old female with a history of repeated episodes of ballottement in lower abdomen few months back for which she consulted some registered medical officer in the peripheral health centre and was advised USG scanning which revealed a right adnexal mass of about 8 × 6 cm. Patient left further investigation as such as she was symptomatically stable. After around 1 year, she started with similar symptoms which actually were on and off. She was referred to tertiary center and advised USG scanning which read as right adnexal mass of 11 × 8 cm. Rest routine investigations were done which includes TLC, DLC, and hemoglobin. Hemoglobin was 9.8 gm/dL, and TLC is 7,800/mL (with eosinophils 38%). Other investigations including enzyme-linked immunosorbent assay (ELISA), coagulation latex agglutination, countercurrent immunoelectrophoresis (CCEP), and CT were not done because of the nonavailability and financial constraints.

There was no history of fever, itching, and weight loss present. The patient was later operated (laparotomy followed by cystectomy), and cystectomy specimen was sent for histopathological examination. Grossly, the cyst measured 11 × 8 cm, uniloculated, and whitish in color with papery thin cyst wall. Cut section showed white gelatinous appearance of membrane which at places was pearly white. Histopathological examination showed numerous fragments of laminated acellular ectocyst along with fibrous pericyst and inner germinal layer (hydatid cyst) (Fig. 2).

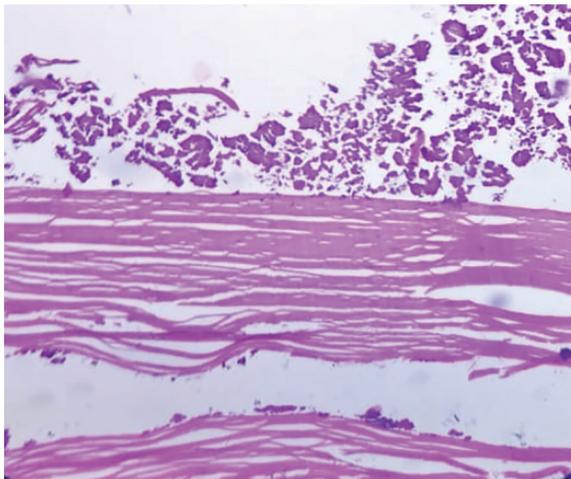
**Case 3**

A 34-year-old female presented to the outpatient department with complaints of dull aching pain in the left lower abdomen for 3 months and fever on and off. On general physical examination, patient appeared to be anemic and was febrile. No history of burning micturition present. She had a previous history of operation for subhepatic hydatid cyst about 5 years back. On clinical examination, abdomen appeared distended and dull mass could be palpated in the left lower abdomen. There was no hepatosplenomegaly and ascites in the abdomen. Routine investigations were done which showed mild anemia (hemoglobin, 9.2 gm/dL), routine urine was normal, and mild raised leukocyte count (14,000/dL) with mild eosinophilia on differential leukocyte

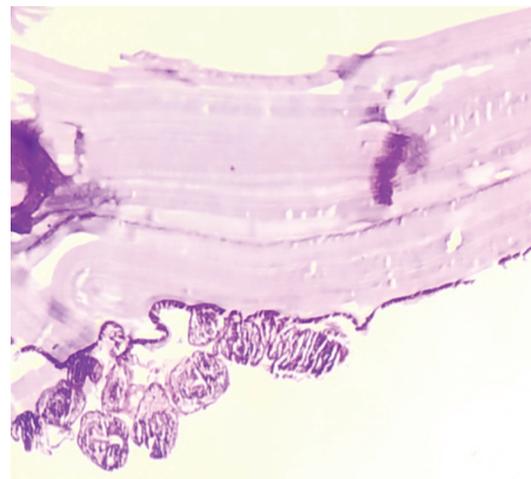
count (neutrophils 50, lymphocytes 36%, monocyte 6%, eosinophils 9%). ESR was 21 mm in first hour. C-reactive protein was 8 mg. Kidney and liver tests were normal. Ultrasonography (USG) of the abdomen and pelvis was done which revealed a cystic lesion of approximately 7 × 5 × 4 cm on the left ovary (left ovarian cyst). Exploratory laparotomy with excision of whole ovary with cyst was done. Specimen was then sent to the Department of Pathology for histopathological examination. On gross examination, cyst measured 7 × 5 × 4 cm, papery thin with glistening cyst wall present. On cut section, thick mucoid material oozed out with pearly white areas present inside the cyst wall. Microscopic examination of the cyst fluid revealed scolices, brood capsules, and hooklets. Histopathological examination of hematoxylin and eosin (H&E)-stained section revealed the inner germinal layer, laminated hyaline ectocyst, and outer pericyst which confirmed hydatid cyst. After confirmation of hydatid cyst, the patient was placed on albendazole therapy of 400 mg twice a day for 4 weeks and advised for follow-up (Fig. 3).

**Case 4**

A 42-year-old female presented to the outpatient department with complaints of right upper quadrant pain with dyspepsia, nausea, and occasional episodes of vomiting. There was no history of jaundice or fever. Physical examination revealed mild tenderness in the right upper quadrant of the abdomen. Murphy sign was positive. Routine blood investigation was done showing total leukocyte count (12,000/cumm with mild eosinophilia in differential leukocyte count). Liver and kidney function tests were normal. USG of the abdomen revealed cystic lesion in the right lobe of liver. Computed tomography (CT) of the abdomen showed well-defined cystic lesion with internal septations and septal and peripheral calcifications in the segments of IVa/IVb of the liver. The gallbladder was distended with hyperdense calculi or mass lesion detected, suspecting to be a hepatic hydatid cyst which may abutting the gallbladder. USG FNAC of the cystic lesion was planned which showed very scant cellularity, few fragments of lamellar teguments from cyst wall along with scattered parasitic hooklets, and calcareous corpuscles in a necrotic inflammatory background. Right subcostal laparoscopy with extended cholecystectomy was planned. Laparoscopy



**Fig. 2:** Hematoxylin and eosin, 40X magnification with typical section through cyst wall layers of echinococcosis showing outer fibrous wall, the laminated layer, and the inner germinal epithelial layer



**Fig. 3:** Microphotograph of hydatid cyst shows protoscolices (hematoxylin and eosin 40X)

revealed a primary cyst of gallbladder which was distended; appeared oedematous, inflamed, and embedded in the hepatic bed. Complete pericystectomy with cholecystectomy followed. The histopathological examination later confirmed the presence of calcified hydatid cyst of gallbladder which was developed entirely extramucosally. Patient was then started with albendazole tab 400 mg BD for 4 weeks followed by a 2-week treatment free interval cycle in three cycles for 3 months (Figs 4 and 5).

#### Case 5

A 45-year-old male presented with epigastric pain for last 4 months. On physical examination, no jaundice was evident. An abdominal lump measuring 5 × 4 cm occupying the epigastric and left hypochondrial region was noted. USG and CT revealed similar findings showing heterogeneous area of 5 cm in size in the body of the pancreas. Liver function test were normal in range, but elevation of C-reactive protein value (15 mg/dL) and amylase (698 U/L) was there. Primary diagnosis of pancreatic pseudocyst with pancreatitis was considered, and surgical intervention was planned after a course of antibiotics for few days. After surgical exploration, cystectomy was done which was sent for histopathological examination. Histopathology biopsy revealed hydatid cyst.

## DISCUSSION

Hydatid cyst disease is a cyclo-zoonotic disease where the causative organism is *Echinococcus granulosus* belong to the family cestodes (tapeworm). Out of two common genus which include *Echinococcus granulosus* and *Echinococcus multilocularis*, the unilocular cyst is mainly caused by *Echinococcus granulosus*.<sup>1</sup> The definite hosts are dogs, foxes, and wolves, while

intermediate hosts are sheep, cattle, and horses.<sup>2</sup> Human is an accidental host and does not play a role in the biological cycle of worm. The mode of infection is by the ingestion of food contaminated with dog feces and also by direct contact with dogs. The ingestion by humans of such contaminated food leads to hatching of the ova in the gastrointestinal tract. The enclosed embryos are liberated in the duodenum and transported to the liver by portal circulation. The liver acts as 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 case. Some escape from these filters and develop in other organs. The incubation period is highly variable. The cyst grows at a rate of 0.3–1 cm/year and may take 5–20 years to attain sufficient size to cause symptoms.<sup>3</sup> It is commonly found in the temperate zones of the world in endemic countries India, Africa, Middle East countries, South America, New Zealand, and China where sheep rearing is a common practice. Endemicity in India is only because of the lack of access to clean potable water supplies and a close association of people with domestic animals like sheep and dogs.<sup>4</sup> The most common sites affected are liver (63%) and the lung (25%), followed by muscles (15%), bones (3%), kidney (2%), spleen (1%), and other sites (1%).

A primary subcutaneous location is extremely rare entity, even in countries where the *Echinococcus* infestation is endemic. The mechanism of primary subcutaneous localization of hydatid cyst is still not clear. The ingested parasite larvae penetrate the intestinal wall, enter circulation, and via portal vein reach the liver. Few may pass through the liver which is said to be first filter and reach the lung, said to be second filter, and reach systemic circulation. Dissemination through lymphatic channels has also been suggested as a possible mechanism and may account of solitary cyst in uncommon site.<sup>5</sup>

Pelvic hydatid disease as well as hydatid cyst of ovary is uncommon sites, and the reported incidence lies in the range of 0.2–2.25%.<sup>6,7</sup> Primary pelvic hydatid cyst's pathophysiology is not clearly established. There are theories that, in such cases, may be the hydatid embryo gains entry to pelvic either by hematogenous or lymphatic route.<sup>8</sup> No specific clinical feature of pelvic hydatid cyst is there, but often presents as an unusual pelvic mass which in case of female as in our Case 2 suspected to be an adnexal mass lesion. First line of investigation is always ultrasonography. But CECT pelvis gives more precise information regarding size location, adherence to neighboring structures, and excellent depiction of the visceral organ involvement.<sup>9</sup> The common sites of hydatid cyst in female reproductive tract are pouch of Douglas and the uterine cavity, but the cyst arising from the ovary is extremely rare.<sup>10</sup> Pelvic echinococcosis symptomatology is nonspecific and includes abdominal pain, dull ballottement of lower abdomen, menstruation irregularities, infertility, and urinary disturbance.<sup>11</sup> Hydatid cyst can simulate either polycystic disease or malignancy, and so difficulties for making correct diagnosis arise due to nonspecific clinical symptoms associated with atypical ultrasonographic and radiological findings which merely show a solid ovarian mass or a benign ovarian cyst.<sup>12</sup> CT scan confirms the diagnosis revealing daughter cysts and contingent calcifications of the cyst wall. Thus, it is important to keep in mind this rare entity as a differential in patients presenting with cyst wall at ovary, causing mass effect, mainly when preceding from an endemic area.

The liver (70–80%) and lungs (15–25%) are the most frequent locations for echinococcal cyst, while primary hydatid cyst of gallbladder is an extremely rare entity.<sup>13</sup> Few case report of gallbladder daughter cysts secondary to liver cyst have been published.<sup>14</sup>



Fig. 4: Gross examination of gallbladder cyst component

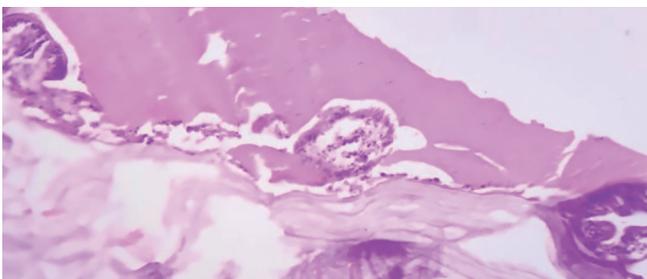


Fig. 5: Histopathology of hydatid cyst showing brood capsule containing viable scoleces (H&E 40X)

Because of the small number of cases reported, the pathogenesis of primary gallbladder hydatid cysts has been opinioned depending upon the location of the cyst whether located in the lumen or in the external surface. If the cyst was found inside the gallbladder, then it was emphasized that it is the result of brood capsules dissemination through the biliary tract, i.e., through cystic duct.<sup>15</sup> Our Case 4 present with primary gallbladder hydatid cyst which was larger in size and laparotomy showed very clearly that the hydatid cyst arises from the gallbladder, grown entirely extramucosally and finally microscopy also confirmed. So in our case of primary gallbladder cyst, transport of oncospheres from the intestine to the gallbladder is more likely to have occurred by lymphatic circulation.<sup>16</sup> Our case present with little difficulty in diagnosing the site of origin of cyst. Even the USG and CT do not clearly indicate the site of origin. In such cases, surgical exploration of cyst with histopathological examination is the only confirmatory method left.

Pancreatic hydatid is a rare entity, incidence ranging from 0.1 to 2%.<sup>17</sup> Most common mode of spread is hematogenous. Cysts in pancreas can present as obstructive jaundice. Cyst in body and tail is usually asymptomatic; findings are similar to our Case 5 with no history of jaundice and cyst present in the body. The possible modes of spread of cystic elements to pancreas are described through biliary system, lymphatic spread from the intestinal mucosa direct passage via pancreatic veins, and retroperitoneal dissemination.<sup>18</sup> Clinical presentation depends on the location of cyst. Cyst located in head of pancreas can present as obstructive jaundice due to external compression of common bile duct.<sup>19</sup> Cyst located in body and tail of pancreas is usually asymptomatic until they become large enough to present as an abdominal lump or cause some symptoms due to compression of adjacent structures like epigastric pain, nausea, and vomiting.<sup>17</sup> The diagnosis of pancreatic cyst can be performed by ultrasonography, CT scan, and MRI. The radiological imaging features are helpful for distinguishing hydatid cyst from other cystic lesions. Even with fairly characteristics features, it is still a diagnostic challenge as most of the time it was reported as pancreatic pseudocyst or cystic neoplasm.<sup>20</sup>

## CONCLUSION

In this paper, we described five cases of hydatid cyst occurring at unusual sites. Hydatid cyst is more common in females, and eosinophilia is associated with most of the cases.

*Echinococcus* should be considered in the differential diagnosis of any cystic lesions in any anatomic location, with or without viscera involvement particularly in endemic areas.

An ovarian hydatid cyst is rare finding. On USG, it mostly mimics cystic ovarian disease or solid ovarian mass. High degree of suspicion combined with USG and CT is required to make preoperative diagnosis. Gallbladder primary hydatid cyst has different spread route of parasite embryos and a better prognosis due to earlier manifestation of symptoms leading to earlier treatment.

Pancreatic head cyst can masquerade as pseudocyst or cystic neoplasm of pancreas. It should always be considered in differential diagnosis of cystic pancreatic lesion especially in patients from endemic regions. Radiological findings together with histopathological examination remain the main stream of investigations for confirmatory diagnosis of hydatid cyst.

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