# Three Years Study Of Unusual Hydatid Cysts: From Head To Toe

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## **ABSTRACT**

Hydatidosis is a zoonotic infection due to larval stage of the tapeworm echinococcus. In adults' liver represents the prevalent site and lungs are the second commonest site. Although these are common sites, it can occur at any site including spleen, pancreas, peritoneum, retroperitoneum, central nervous system, soft tissues, and breast. We present the histopathological spectrum of hydatid cyst at unusual sites in our hospital over a period of three years. The present retrospective study was conducted in the department of Pathology, SKIMS during a period of 3 years from July 2019 to June 2022 where a total of 12cases were diagnosed as Hydatid Cyst at unusual sites on HPE during study period were included. Male and female ratio of incidence found to be 1:1:4. Maximum no of cases i.e., 3 occurred in brain, 2 in spleen,2 in thigh,1 each in breast, omentum, kidney, sacral ala and mediastinum. The hydatid cyst can be present in any part of the body and there is no natural immunity that inhibit the development & growth of it. These unusual locations frequently result in nonspecific symptoms. To avoid lifethreatening complications and unnecessary radical surgeries, hydatid cystic disease should always be suspected in all cystic lesions of radio imaging investigations, especially in endemic areas.

Key Words: Hydatid cyst, CBD, HPE.

# **INTRODUCTION:**

Hydatid disease or hydatidosis (HD) is a Greek term which is caused by the larval stage of the ribbon helminths of parasite Echinococcus which causes deformity of internal organs (liver, lungs heart & brain). According to WHO, infectious and parasite diseases, which account for 16 million deaths worldwide each year, are still the primary causes of human mortality in the 21<sup>st</sup> century<sup>1</sup>. The most common form in humans is caused by E. granulosus. Infestation with larval stages of dog tapeworms can result in encystation in various organs commonly involved organs include the liver (75%) and lungs (15%), while the remaining 10% occurs in other body organs including spleen, kidney, pancreas, peritoneum, retroperitoneum, central

nervous system, soft tissues, and the breast [1-2]. Theoretically, it can occur at any site except teeth, hair, and nails [3]. A possible dissemination through lymphatic channels accounts for cases with hydatid cysts at uncommon sites [4,5]. The exact percentage of site involvement varies and the exact incidence of unusual locations is difficult to ascertain as they are only reported as case reports<sup>6</sup>. Breast, is a rare site of involvement accounting for only 0.27% of all cases of Hydatid disease. [7,8,9]. Cerebral involvement is very rare (1-3%), and more common in children [10]. Hydatid cysts in the cerebral are often supratentorial, infratentorial lesions are extremely uncommon. Intracranial hydatid cysts are commonly solitary. Multiple intracranial cysts are rare [11]. Incidence of hydatid disease of bone from various studies is reported to be 0.5-4% [12]. About 60% cases of bone hydatidosis affect the spine and pelvis, 28% the long bone and 8% the ribs and scapula [13]. Skeletal lesions in hydatidosis tend to present with pain or pathological fractures following trivial injuries [14].

# **MATERIALS AND METHODS**

The present study was conducted in the department of Pathology, SKIMS. In this study patients attending department of Pathology, SKIMS from July 2019 to June 2022 were included. A written informed consent was also taken from the cases. In this retrospective study the patients having hydatid cyst at unusual sites like kidney, spleen, retroperitoneum, brain, bone and omentum were included in the study. Specimens received were fixed in formalin; HP diagnosis was made on routine H&E sections. The diagnosis was based on the history, physical examination, laboratory tests, and radiographic examinations in all patients. Six patients underwent ultrasonography, and all underwent computed tomography (CT) imaging.

## **RESULTS**

A total no. of 12 cases which were diagnosed as Hydatid Cyst at unusual sites on HPE during study period were included. The youngest patient reported was 7years old, while the oldest patient reported was 60yrs. Out of 12 cases 5 were males & 7 females. The hydatid cyst ratio of incidence in Male and female was found to be 1:1.4. Maximum no of cases i.e., 3 occurred in

brain, 2 in spleen, 2 in thigh, 1 each in breast, omentum, kidney, sacral ala, omentum and mediastinum.

Table 1. Age incidence in cases

Age group	No. of cases
0 to 10 years	2
11 to 20 years	2
21 to 30 years	3
31 to 40 years	1
41 to 50 years	1
51 to 60 years	3

Table 2. Incidence of cases at unusual sites

Site	No. of cases
Brain	3
Spleen	2
Thigh	2
Breast	1
Omentum	1
Kidney	1
Sacral Ala	1
Anterior Mediastinum	1

**Table - 3: Clinical findings** 

Case	Clinical presentation
1.Brain 11y, Male	Headache
2.Brain 7y, Female	Weakness on right side of body
3.Brain 7y, Female	Recurrent headache and Right hemiparesis
4.Spleen 21y, Male	Pain abdomen
5.Spleen 60y, male	Jaundice
6.Thigh 40y, male	Thigh abscess
7. Thigh 30y, male	Pain and swelling left thigh

8.Kidney 53y, female	Pain right lumbar region
9.Breast, 45y, female	Swelling in the right breast associated with mild pain
10.Sacral Ala,16y female	Mild Pain sacral area
11.Anterior Mediastinum,25y, female	Breathlessness
12.omentum,53y, male	Pain abdomen

Case	Lab./ radiological findings/ intraoperative findings
1.Brain 11y, Male	Huge Cyst with daughter cysts involving almost whole of the left cerebral hemisphere.
2.Brain 7y, Female	Cyst involving left motor cortex
3.Brain 7y, Female	Heterogeneously enhancing three cysts in the left temporal lobe
	Three big endocysts present in the left temporal lobe
4.Spleen 21y, Male	Case of disseminated Hydatid disease
5.Spleen 60y, male	Mid CBD growth with splenic hydatid cyst and cholelithiasis
6.Thigh 40y, male	3x2cm collection in right thigh
7. Thigh 30y, male	Non enhancing cystic lesion measuring 10x6 cms.
	Serology for hydatid was positive
8.Kidney 53y, female	8x6 cm cystic lesion right kidney
9.Breast, 45y, female	USG breast revealed a thick-walled round to oval complex infected cystic lesion
	measuring 69x61x54 mm in the outer quadrant of right breast with internal
	septations and associated inflammatory changes in the adjacent breast
	parenchyma
10.Sacral Ala,16y female	Case of recurrent hydatid cyst
	IOF: Large hydatid cyst within right sacral Ala with multiple daughter cysts
11.Anterior Mediastinum,25y, female	Non enhancing cystic lesion measuring 10x8 cms.
12.omentum,53y, male	Infected ruptured omental hydatid cyst with hemoperitoneum

Case	Morphology
1.Brain 11y, Male	Gross; whitish membranes measuring 18x8cms.HPE; Laminated membranes a with many Scolices
2.Brain 7y, Female	Gross; whitish membranes measuring 14x4 cms.HPE; Scolices and Laminated membranes
3.Brain 7y, Female	Gross; Three pearly white cystic structures measuring 5x4cms;3.5x1.5 cms and 3.5x3.5 cms HPE; Laminated membranes consistent with hydatid cyst
4.Spleen 21y, Male	Gross; Splenectomy specimen measuring 10x5x5, weighing 150 gm. External surface showing a cyst measuring 4x3x2cms.On cutting open cyst whitish membranes taken out along with daughter cysts. HPE; Laminated membranes along with Inflammatory infiltrate
5.Spleen 60y, male	Gross; Splenectomy specimen measuring 13x10x6, weighing 250 gms. Capsular breach identified by cystic structure. Cut section shows two cystic structures filled with membranes.  HPE: Laminated membranes consistent with hydatid cyst
6.Thigh 40y, male	Gross: single soft tissue piece measuring 2.5x2cm. External surface is unremarkable. Cut section is grey white HPE: Laminated membranes of hydatid cyst against inflammatory background and necrosis
7. Thigh 30y, male	Gross: Well encapsulated globular cystic structure measuring 10x5x3cm.On cutting open pearly white membranes filled with thick yellowish membranes identified HPE: Laminated membranes of hydatid cyst
8.Kidney 53y, female	Gross:8 pearly white membranes altogether measuring 8x5cms.No normal renal parenchyma identified HPE: Laminated membranes of hydatid cyst
9.Breast, 45y, female	Gross: The cyst was pearly white in color measuring 7x7 cms and the inner surface was covered with sand like particles. HPE: laminated membrane of hydatid cyst with chronic inflammatory infiltrate rich in eosinophils in the surrounding fibro collagenous tissue.
10.Sacral Ala,16y female	whitish laminated membranes of hydatid cyst
11.Anterior Mediastinum,25y,female	Gross: Globular cystic structure 10x3.5 cms,30ml clear fluid drained out HPE: revealed laminated membrane of hydatid cyst with surrounding pericyst showing chronic inflammation
12.omentum,53y, male	Glistening whitish laminated membranes of hydatid cyst

# **DISCUSSION**:

Hydatid disease is a parasitic infection caused by the larval form of Echinococcus granulosus & it is endemic among sheepraising communities, particularly in regions of South America, the Mediterranean shore (Spain, France and Italy), Eastern Europe, Turkey, East Africa, Central Asia, China, and Russia [15]. The prevalence of HC shows regional differences and ranges between 0-79 per 100.000 population<sup>[16]</sup>. The state of Kashmir, India, is endemic for hydatid disease<sup>[17]</sup>. Echinococcus granulosus (EG) is a 5 mm long hermaphroditic tapeworm [18,19]. The adult E. granulosus produces eggs that are released in the stool of infected canines. Eggs ingested by intermediate hosts (cows, sheep and humans) release embryos in the duodenum, which penetrate the intestinal mucosa and enter into circulation [20]. The liver acts as the first filter, while the lungs act as a second filter. Only 15% of the embryos are free to develop cysts in other organs of the body [21]. Thus, the liver is the most common site affected (75%), followed by lungs (15%), muscles (4%), kidney (2%), spleen (2%), bone (1%) etc [22].

Hydatid cysts of the breast are extremely rare even in endemic areas, accounting for only 0.27% of all cases [23]. Very few cases of hydatid cysts of the breast have been reported in the literature and the largest series of 20 hydatid cyst of the breast was reported in Tunisia [24]. The breast can be a primary site of infection or part of a disseminated hydatidosis [23]. Clinically, a hydatid cyst of breast usually presents with a painless, slowly increasing lump in the breast, of long duration without axillary lymphadenopathy. It affects generally women in the age group of 30-50 years [22]. Ultrasonography and mammography are very effective in the evaluation of this mass. Despite its high cost, MR imaging has also been used in further evaluation of mass. Serological investigations – indirect hemagglutination test, may be used for diagnosis and in the follow-up of patient [22]. Preoperative diagnosis can be made by fine needle aspiration cytology (scoliosis, hooklets or laminated membrane can be identified), but the use of fine needle aspiration is controversial. There are only a few studies describing this method without complications [21,26], but puncturing of the cyst may lead to an anaphylactic reaction and secondary cyst development due to spillage of hydatid fluid [27]. Our case was 45-year-old female, with swelling in the right

breast with mild pain. On examination there was swelling about 3x2 cms, soft to cystic in consistency. FNAC was done which yielded 60 ml of clear fluid and a diagnosis of fibrocystic changes of the breast was made on microscopy's breast revealed a thick-walled round to oval complex infected cystic lesion measuring 69x61x54 mm in the outer quadrant of right breast with internal septations and associated inflammatory changes in the adjacent breast parenchyma. The cyst along with the adjacent inflammatory tissue (pericyst) were removed subsequently. On histopathological examination of the specimen the laminated membrane of hydatid cyst was seen with chronic inflammatory infiltrate rich in eosinophils in the surrounding fibro collagenous tissue.

Cerebral involvement is very rare (1-3%), and more common in children [10]. Cerebral hydatid cysts are usually supratentorial, the infratentorial lesions are quite rare. Intracranial hydatid cysts are commonly solitary. Multiple intracranial cysts are rare [11]. Patients with intracranial hydatid cysts usually present with focal neurological deficit and features of raised intracranial pressure; the latter may be due to the large size or due to interference with pathway of CSF. The typical intracranial hydatid cysts caused by Echinococcus granulosus, present as a well-defined solitary cystic lesion in the middle cerebral artery territory in parietal lobes, although they can be seen in any location including skull vault, extradural, intraventricular, meningeal, posterior fossa and brainstem [28]. Operative diagnosis of hydatid cysts can be made by USG and confirmed by a CT scan. The magnetic resonance imaging is also of considerable value in intracranial hydatidosis. Surgically, intact cyst excision is the ideal treatment. Medical treatment with albendazole seems to be beneficial both pre-and postoperatively [11,29]. The definitive diagnosis can be made by histopathologic examination [30]. All the three cases in our study were children with cerebral involvement. One case had multiple (three) cysts. Pre operative diagnosis was made on MRI. Surgically intact cysts were excised and histopathology was consistent with hydatid cyst. Incidence of hydatid disease of bone from various studies is reported to be0.5-4% [12].

Primary isolated bone hydatid is a very rare occurrence. The lesions in bone may lie dormant for 10 to 20years [31]. Spine is the common site of infection [32]. Hydatid disease of spine usually spreads over the spine by direct extension from pulmonary, abdominal or pelvic infestation and most commonly affects the thoracic (52%), followed by the lumbar (37%) and then the cervical and sacral sacral spine [33]. Skeletal lesions in hydatidosis tend to present with pain or pathological fractures following trivial injuries [14]. The most common radiological manifestation of skeletal hydatid disease is a lucent expansile lesion with cortical thinning. Bone hydatid disease lacks a typical clinical appearance and image characteristics on X-ray or CT scan are similar to those of tuberculosis, metastases, giant cell tumor or bone cysts. [34]. Magnetic resonance imaging shows distinctive diagnostic features of bone hydatid disease, especially in the spine. The only definitive treatment when bone is involved is complete resection of the involved area with a wide healthy margin. The combination of anthelminthic therapy, wide resection and the use of polymethylmethacrylate (PMMA) gives the best outcome in the treatment of bone hydatidosis [33]. Our case was the young 16year-old female with mild pain in the sacral region. She was already operated in the past for hydatid cyst liver. Intraoperative findings revealed large cyst with daughter cysts in the sacral ala area. HPE revealed laminated membranes of hydatid cyst.

The involvement of spleen in HC is rare. It is the 3rd most common site and the prevalence of splenic involvement ranges between 0.9% and 8% [35,36,37]. Splenic HC generally develops by means of systemic dissemination or intraperitoneal spread from a ruptured liver cyst. Isolated splenic involvement is very infrequent [38] and splenic HCs are generally solitary [38]. splenic hydatidosis should be differentiated from other splenic cystic lesions, such as epidermoid cyst, abscess, hematoma, post traumatic pseudocyst, neoplasms like lymphangioma and hemangioma [39,40]. We had two cases of splenic hydatid cyst. One patient was young 21-year-old male with disseminated hydatid disease. Second patient was elderly male 60 years and was being evaluated for CBD growth. On HPE he was found to be having two hydatid cysts in spleen along with

Adenocarcinoma of Gall bladder in the background of Xanthogranulomatous cholecystitis.

Soft-tissue HC occurs in 0.5-4.7% of patients living in endemic areas <sup>[38,41]</sup>, the growth of the cyst within a muscle is difficult due to the contractility of muscles and presence of lactic acid <sup>[38,36]</sup>. HC has an affinity for muscles of the neck, trunk and limbs because of increased vascularity and decreased activity of these muscle groups <sup>[38,36]</sup>. soft tissue HCs are often confused with benign soft tissue tumors <sup>[36]</sup>. We had two cases of soft tissue hydatid both in the thigh. One case presented as thigh abscess. Other case had radiology suggestive of hydatid with positive hydatid serology.

Renal involvement is rare (1-4%) [38,36]. It is reported as the common site following liver and lung in several articles [42]. they are mostly solitary and located at upper pole or cortex [38]. Multilocular HCs can be misdiagnosed as simple renal cysts, cystic nephroma, and cystic variants of renal cell carcinoma [38] and infected HCs can be misdiagnosed as renal abscess [38]. The mediastinal hydatid cyst is uncommon but it should be included in the differential diagnosis of the mediastinal cyst in endemic parts of the world [43]. The omental and retroperitoneal hydatid cysts are very uncommon, but these cysts can become huge in size [44].

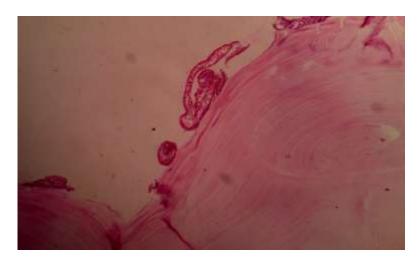
# **CONCLUSION**

The hydatid cyst can be present in any part of the body and there is no natural immunity that inhibit the development & growth of it. These unusual locations frequently result in nonspecific symptoms. To avoid life-threatening complications and unnecessary radical surgeries, hydatid cystic disease should always be suspected in all cystic lesions of radio imaging investigations, especially in endemic areas.

# **FIGURE LEGENDS**



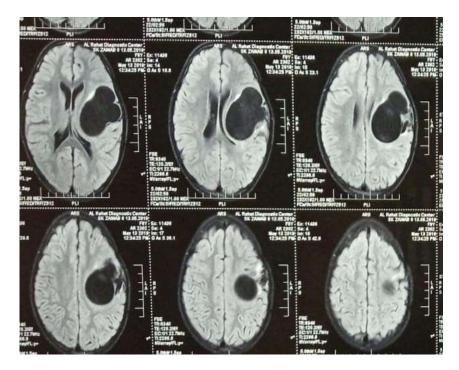
**Fig 1** USG breast revealing a thick-walled round to oval complex infected cystic lesion in right breast with internal septations and associated inflammatory changes.



**Fig2.** Gross picture of the cyst pearly white in colour measuring 7x7 cm and the inner surface covered with sand like particles in breast hydatid.



**Fig 3** Histopathological examination of the specimen showing laminated membrane of hydatid cyst seen with chronic inflammatory infiltrate rich in eosinophils in the surrounding fibrocollagenous tissue in breast hydatid



**Fig 4** MRI brain revealing heterogenous enhancing three cysts within left temporal lobe suggestive of hydatid cyst.



**Fig 5** Grossly received three pearly white cystic walls measuring as 5x4cm,2.5x2.5cm and 2.5x1.5 cm.

# **CONFLICTS OF INTEREST:**

The author(s) declare(s) that there is no conflict of interest regarding the publication of this paper.

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