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# CASE REPORT IMAGING IN "PELVIC HYDATIDOSIS" "AN UNUSUAL ECHINOCOCCOSIS"

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## ABSTRACT

Hydatidosis is a parasitic infestation caused most commonly by *Echinococcus granulosus* and may occur in any organ or tissue. The most common site involved is liver (59–75%), followed in frequency by lung (27%), kidney (3%), bone (1–4%), and brain (1–2%). Other sites such as the heart, spleen, pancreas, omentum, ovaries, parametrium, pelvis, thyroid, orbit or retroperitoneum, and muscles are very rarely affected. Primary hydatid cyst in rectovesical pouch is an extremely unusual condition. Pelvic Hydatidosis is a rare entity. Here, we present a Caseof Pelvic Hydatidosis with characteristic clinical and imaging findings- a seventeen year old young male presented to the Department of Casualty, Mahatma Gandhi Medical College and Hospital, Jaipur (Rajasthan) with the Complaints of pain in lower abdomen that was dull, intermittent, non-radiating with no relation to food or any other specific activity and burning micturition. Ultrasound & CT scan of abdomen showed hydatid cyst in recto-vesical pouch. Surgery was performed and pathology of the excised mass confirmed the diagnosis. This case of primary hydatid cyst in recto-vesical pouch is being presented because of its rarity.

*Keywords: Pelvic Hydatid Cyst, Echinococcus Granulosus, Recto-Vesical Pouch, U.S.G. – Ultrasonography, C.T. – Computerized Tomography* 

## **INTRODUCTION**

Hydatid disease or echinococcosis is a cyclo-zoonotic parasitic disease caused by infection with larva (metacestode) of the cestode Echinococcus. Four species of the genus echinococcus are known to cause infection in humans; *Echinococcus granulosus* (cystic hydatid disease), *Echinococcus multilocularis* (alveolar hydatiddisease), *Echinococcus vogeli*, and *Echinococcus oligarthus* (both causing polycystic hydatid disease) (Khuroo, 2002). *Echinococcus granulosus* requires two hosts. Dog is the definite host of echinococcus granulosus and man can become the intermediate host through contact with infected dogs or by ingesting contaminated food. As liver and lungs act as first and second filter respectively for the embryos, which are released after ingestion of ova, therefore very few embryos enter the systemic circulation. At the site of deposition, the embryo develops into bladder or cyst filled with fluid and is called hydatid cyst (Greek hydatisa drop of water).

Although liver is the most common site of echinococcal involvement, yet cystic echinococcus infestation can occur in any part of the body and should be considered in the differential diagnosis of cystic masses (Engin *et al.*, 2000). Peritoneal hydatidosis occurs in 12% of cases and is usually the result of traumatic or surgical rupture of a hepatic or splenic cyst (Gossior *et al.*, 1997). Primary peritoneal hydatid cyst is rare and the mechanism of primary peritoneal infection by the parasite is still unclear (Gelati *et al.*, 2003; Mansori *et al.*, 2000). Implantation of the hydatid larvae in such cases could be haematogenous. In our case the hydatid cyst found in rectovesical pouch was primary as there was no such cyst in any other organ, thereby making it an extremely rare condition. No specific reference was found in literature about primary hydatid cyst in rectovesical pouch although there are few references of primary hydatid cysts which were retrovesical (Ben Adbullah *et al.*, 2000; Horchani *et al.*, 2001; Moussaoiri *et al.*, 1994; Tocchi *et al.*, 1999). Ultrasound & CT scan are helpful in diagnosis besides clinical presentation (Gossior *et al.*, 1997; Kotoulas *et al.*, 1990). Symptomatic large hydatid cysts should be treated surgically and cystopericystectomy remains the gold standard procedure (Balik *et al.*, 2001). The surgical approach used

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in our case was conservative (partial pericystectomy) because of the site and the anatomopathologic characters of the cyst.

#### CASES

A seventeen-year-old young male presented to the Department of Casualty, Mahatma Gandhi Medical College and Hospital, Jaipur (Rajasthan) with the Complaints of pain in lower abdomen. Pain was dull, intermittent, non-radiating with no relation to food or any other specific activity. The patient also complained of burning micturition. Bowel habits were normal.

There was no history of trauma or surgery in the past.

There was no relevant past or family history.

### Physical and Clinical Examination

Patient was a young male, moderately built and nourished& well-oriented to time, place and person.

His general physical examination and vitals were normal. He did not have anemia, icterus or lymphadenopathy. Liver and spleen were not palpable & there was no ascites on per abdomen examination. There was a well-defined palpable lump in pelvic region, cystic in feel, immobile, non-tender with normal local temperature. Examination of other systems was unremarkable.

Routine biochemical and haemotological investigations were normal except differential leucocyte count & routine examination of urine showed eosinophilia and urinary tract infection, respectively.

### Radiological Examination

*Ultra Sonography of Abdomen* showed a multiloculated cystic lesion measuring 47 mm x 43mm x 40 mm in recto-vesical pouch, indenting rectum posteriorly and urinary bladder anteriorly. The wall & the septae of the cyst were hyper echoic giving the appearance of cysts (daughter) with in a cyst, which is pathognomonic of hydatid cyst. No such cystic mass was seen in liver, spleen, kidneys or any other abdominal organ.

*CT* Scan of Abdomen revealed a well-defined, thick walled, multi-loculatedcystic lesion measuring 4.7cms x 4.3cms x 4.0cms in left side of pelvis situated in left peri-rectal region posterior to left seminal vesicle. The internal portion of lesion showed mild non-enhancing hypo-attenuation. The wall was thick and showed mild contrast enhancement. No air-fluid level was seen. Small daughter cysts were seen within the dependent portion of the lesion. All other abdominal organs were normal.

*Surgery* was performed to excise the mass. Although pericyst was involving the posterior wall of urinary bladder, yet meticulous dissection was done to separate the cyst wall & take out the mass intact, thereby preventing the spillage of contents. Some of the pericyst was left behind in order to preserve the wall of urinary bladder. Postoperatively patient was given albendazole for one month.

On macroscopic examination the mass was cystic, pearly white in colour with opaque wall. The wall of the cyst was cut open and it revealed multiple small cysts (daughter cysts). The small cysts contained hydatid fluid with hydatid sand, which represented the scolices. The microscopic examination revealed laminated membrane & scolices.

#### DISCUSSION

Hydatid disease or echinococcosis is a cyclo-zoonotic parasitic disease caused by infection with larva (metacestode) of the cestode Echinococcus. Four species of the genus echinococcus are known to cause infection in humans: Echinococcus granulosus (cystic hydatiddisease). *Echinococcus* multilocularis(alveolar hydatid disease), Echinococcus vogeli, and Echinococcusoligarthus (both causing polycystic hydatid disease) (Khuroo, 2002). Echinococcus granulosus requires two hosts. Humans become accidental intermediate hosts. The most common site involved is the liver (59–75%), followed in frequency by lung (27%), kidney (3%), bone (1–4%), and brain (1–2%). Other sites such as the heart, spleen, pancreas, omentum, ovaries, parametrium, pelvis, thyroid, orbit or retroperitoneum and muscles are very rarely affected (Yuksel et al., 2007).

A typical hydatid cyst is formed from its embryo. It consists of three layers (Milicevic, 1994). The outer layer (pericystor adventitia) consists of fibrous tissue, is grey in color, and blends immediately with the

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liver. It is formed from the host tissue as a result of chronic inflammatory reaction to the parasite. The pericyst usually increases in thickness as the cyst expands. Liver and spleen hydatid cysts have a thick pericyst as compared to peritoneal hydatid cysts, in which the pericyst is extremely thin.

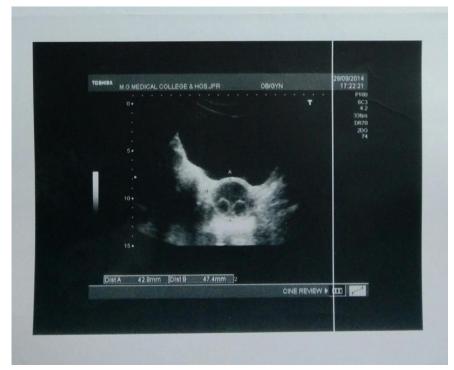


Figure: 1 Ultra Sonographic Image Showing Multiloculated Cystic Lesion – Daughter Cysts with in a Thick Walled Cyst in Recto-Vesical Pouch – Pelvic Hydatid Cyst



Figure: 2 Contrast Enhanced C.T. Axial Image Showing Pelvic Hydatid Cyst with Mild Contrast Enhancement

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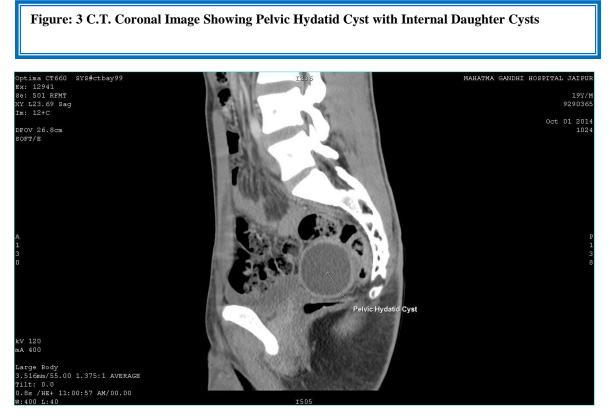


Figure: 4 C.T. Sagittal Image Showing Pelvic Hydatid Cystin Recto-Vesical Pouch

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The hydatid cysts in the lung and brain have no pericyst at all. Complete calcification of the pericyst may interrupt the nutrient and oxygen supply to the parasites, and thus marks the death of the hydatid cyst. It is elastic, made up of gelatinous, chitinous material and when incised or ruptured, curls in on itself, exposing the inner layer. The innermost germinal layer is cellular and consists of a number of nuclei embedded in a protoplasmic mass. It is a very thin, vital layer of the cyst, and produces brood capsules with scolices, secretes hydatid fluid, and forms the outer layer. The cyst fluid is crystal clear and colorless with a specific gravity of 1005–1010, is slightly alkaline, and is highly antigenic and toxic. Contact with the fluid can give rise to anaphylactic shock. Hydatid cysts expand slowly and asymptomatically, and thus tend to be quite large at presentation. The high secretion pressure is responsible for the progressive enlargement of the cyst. Fluid pressure within a hydatid cyst can reach up to 70 cm of water (Milicevic, 1994).

The clinical manifestations of hepatic hydatid disease depend upon the site, size and stage of development of the cyst; on whether the cyst is dead or alive and whether it is secondarily infected or not. As hydatid cysts grow, they can rupture into the surrounding tissues. Internal rupture occurs due to trauma or pressure necrosis from the growing cyst. Unusual complications of a hepatic hydatid cyst include rupture into the stomach, duodenum or small intestine. Rupture into the biliary tree can cause cholangitis, biliary colic and jaundice with excretion of germinative membranes in the stools. Bile-stained cyst fluid is an indicator of communication of the cyst with the biliary tract (Milicevic, 1994). Rupture into the general peritoneal cavity may result in anaphylactic shock and formation of localized or generalized secondary echinococcosis (Dew, 1930).

Peritoneal hydatid cyst, either primary or secondary, represents an uncommon but significant manifestation of the disease (approximately 13%). Intra peritoneal hydatid cysts are usually secondary to the rupture (spontaneous or accidental at surgery) of a primary hepatic, splenic, or mesenteric cyst (Yuksel *et al.*, 2007).

A solitary cyst in the pelvic cavity can be considered primary only when no other cysts are present. In such a case, the hydatid embryo gains access to the pelvis by hematogenous or lymphatic route. Pelvic hydatid cysts usually present as a nonspecific mass with pressure effects on adjacent organs such as the rectum and urinary bladder. Rarely, they can cause obstructed labor, obstructive uropathy, and renal failure. Sometimes, they can rupture spontaneously (Tocchi *et al.*, 1999).

Serology and imaging are the main tools for establishing diagnosis. Ultrasound is the preferred first-line imaging, but contrast enhanced computed tomography gives more precise information regarding the morphology (size, location, neighborhood, and number) of the cyst.

Drug treatment with albendazole has been found to be successful in a proportion of cases, but drug therapy is generally not used as the primary treatment except in cases where the patient is not fit for surgery. Surgery is the most effective treatment. Combination of preoperative albendazole therapy, surgery, and postoperative albendazole therapy (for 6–8 weeks) is a useful regimen. Albendazole suppresses the development of hydatid cysts following intra peritoneal inoculation of protoscolices (Tocchi *et al.*, 1999). In order to prevent the rupture and spillage, meticulous dissection hasto be done and the area surrounding the cyst should be packed with mops soaked in scolicidal agents such as hypertonic saline (20%) and Cetrimide with chlorhexidine (0.1 to 0.5 %) (Parray *et al.*, 2010; Arazi *et al.*, 2005).

## Conclusion

Pelvic echinococcosis is rare, with an incidence of 0.2–2.25%. Pelvic hydatid cysts may have varied and nonspecific presentation. It has to be differentiated from mesenteric cyst. Ultrasonography and computed tomography are both excellent imaging modalities for the detection of hydatid cysts. The treatment of choice for pelvic hydatid cyst is principally a careful and complete surgical excision. The radiologist plays a key pivotal role as imaging is an important tool for early diagnosis and excluding differential diagnoses, which can modify the progression of disease with timely intervention and management.

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#### REFERENCES

Arazi M, Erikoglu M, Odev K, Memik R and Ozdemir M (2005). Primary echinococcus infestation of the bone and muscles. *Clinical Orthopaedics and Related Research* **432** 234-41.

Balik IAA, Celebi F, Basglu M et al., (2001). Intra-abdominal extra-hepatic echinococcus. Surgery Today 31(10) 818-24.

**Ben Adbullah R, Hayri M, Aoun K and Ayed M (2000).** Retrovesical and Retroperitoneal extrarenal hydatid cyst: Descriptive study of 9 cases. *Progrès en Urologie* **10**(3) 424-31.

Dew HR (1930). Some complications of hydatid disease. British Journal of Surgery 18 275-93.

Engin G, Acunar B, Rozanes I and Acunar G (2000). Hydatid disease with unusual localization. *European Radiology* **10**(12) 1904-12.

Gelati G, Fiori E, Sanmartino F *et al.*, (2003). Unusual locazilations of the hydatid cyst: Description of a clinical case. *Minerva Chirurgica* 58 231-34.

Gossior KJ, Kontoyiannio DS, Dascalogiannaki M and Gourtsoyiannis NC (1997). Uncommon locations of hydatid disease: CT appearances. *European Radiology* **7**(8) 1303-08.

Horchani A, Nouira Y, Chtourou M, *et al.* (2001). Retrovesical hydatid disease a clinical study of 27 cases. *European Urology* **40**(6) 655-60.

Khuroo MS (2002). Hydatid disease: Current status and recent advances. *Annals of Saudi Medicine* 22 56-64.

Kotoulas G, Gouliamos A, Kalovidoures A, Vlahos L and Papavasiliou C (1990). Computed Tomographic localization of Pelvic hydatid disease. *European Radiology* 11(1) 38-41.

Mansori O, Zenter A, Sair K, *et al.*, (2000). Peritoneal Hydatid Cyst.Apropos of 12 cases. *Annales de Chirurgie* **125**(4) 351-57.

**Milicevic M** (1994). Hydatid disease. In: Blumgart LH, editor. *Surgery of the Liver and Biliary Tract*. 2nd edition (New York: Churchill Livingstone) 1121-50.

Moussaoiri A, Aboutaieb R, JoualA et al. (1994). Isolated Retrovesical hydatid cyst. Apropos of 2 cases. Journal of Urology (Paree) 100(2) 101-04.

Parray FQ, Ahmad SZ, Sherwani AY, Chowdri NA and Wani KA (2010). Primary paraspinal hydatid cyst: A rare presentation of echinococcosis. *International Journal of Surgery* 8 404-6.

**Perigela HC, Reddy MK, Prasad BV and Narahari J (2013).** Giant pelvic hydatid cyst. *Journal of Dr. NTR University of Health Sciences* **2** 218-21.

Seenu V, Misra MC, Tiwari SC, Jain R and Chandrasekhar C (1994). Primarypelvic hydatidcyst presenting with obstructive uropathy and renal failure. *Postgraduate Medical Journal* 70 930.

Sharma R, Bargotra R, Azad T and Singh M (2004). Hydatid cyst in recto-vesical pouch. JK Science 6(2).

Tocchi A, Mazzoni G, Lepre L et al. (1999). Rare Locations of Hydatid Disease. II Giornale di chirurgia 20(4) 177-80.

Yuksel M, Demirpolat G, Sever A, Bakaris S, Bulbuloglu E and Elmas N (2007). Hydatid disease involving some rare locations in the body: A pictorial essay. *Korean Journal of Radiology* 8 531-40.