

# Sinister Ubiquity of Human Cystic Echinococcosis: A Rare Case of Retro Rectal Hydatid Cyst Presenting as Benign Fistula in Ano.

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**Abstract:** Hydatid disease, a zoonotic infection, is a parasitic affliction caused by *Echinococcus* species. Hydatid cysts by far are usually located in the liver and lung. Retro rectal hydatid is an exceedingly rare occurrence, in comparison. In this case report, we present the case of a 48-year-old male who initially presented to us as benign fistula in ano, and on evaluation, was found to have a retro rectal cystic mass lesion. Magnetic resonance imaging of the pelvis revealed a left postero-pararectal unilocular cyst measuring 8 × 7.8 cm with Type I fistula in ano. The patient successfully underwent enucleation of the cyst, which on histopathological examination proved to be cystic hydatosis. This demonstrates the sinister ubiquity of human hydatid disease in the differential diagnosis of any cystic mass in any anatomic location of human body, and more so in endemic regions.

**Keywords:** General Surgery, Hydatid Disease, Hydatosis, Cystic lesions, Cystic pararectal lesion, North India, Fistula in Ano, Magnetic Resonance Imaging

## 1. Introduction

Hydatid cyst (cystic echinococcosis/CE) is a parasitic infection caused by the larval stage of the cestode, *Echinococcus granulosus* (*E. granulosus*) resulting in the formation of cystic lesions in the infected organs of animal as well as humans hosts. CE has a wide geographic distribution. It is endemic in the Middle East and other parts of the world where livestock is raised in association with dogs. More than 1000, 000 people are affected annually as per the World Health Organization (WHO) (1). Infection occurs by oral ingestion of the infective eggs/ova from the stool of the definitive host such as dogs by the intermediate host such as a human being. The eggs hatch in the small intestine of the host, penetrate the mucosa, enter the portal blood circulation, and then migrate to various organs. It commonly affects the liver and lungs where the hydatid cysts developed are filled with protoscolices and fluid (2). We present a very rare case of retrorectal cyst which was found during the evaluation of a patient who presented a case of fistula in ano. The first case of mesorectal hydatid cyst was published by Lockhart-Mummery et al. in 1935 (3)

## 2. Case Report

A 48-year-old male with no underlying comorbidity presented to our hospital with the main complaint of pain and discharge in perianal area for a duration of past 2 years. There was no history of any previous perianal surgical procedure. The examination of abdomen was normal and perianal examination revealed a simple fistula in ano (Park's I). On digital rectal examination soft lesion was felt in retrorectal space about 5 cm from anal verge and upper end of lesion was not felt. In view of retrorectal mass felt on examination further investigations were ordered. Flexible sigmoidoscopy up to splenic flexure was unremarkable except for a bulge in the posterior wall of upper rectum. Tumor markers were unremarkable and hydatid serology was negative. Chest X-Ray along with

abdominal ultrasonography was unremarkable as well. Magnetic resonance imaging revealed a left posterior-pararectal unilocular cyst measuring 8 × 7.8 cm, hypo intense on T1W images and hyperintense on T2W images, causing indentation of the left postero-lateral wall of bladder with Type I fistula in ano.

Surgery was planned based on the preliminary diagnostic possibility of a cystic nerve sheath tumor, lymphangioma, or hydatid cyst in the order of their respective diagnostic possibilities. During the surgical exploration via a midline incision, a cystic mass was identified on the left side in the mesorectum and the rest of the abdominal cavity did not contain any cyst. Aspiration of the cyst showed clear fluid and a scolical agent was injected into the cyst. The surgical site was irrigated with hypertonic saline (3% NaCl). The laminated membrane was removed as the cyst was adherent to the bladder wall, rectum, and presacral fascia. The surgical site was flushed with hypertonic saline and a drain was placed in the cavity. Postoperative period was uneventful; the drain was removed on 4<sup>th</sup> postoperative day. Patient was discharged on the 5<sup>th</sup> postoperative day with adjuvant therapy of Tab. Albendazole (10 mg/kg/day) for a period of 3 months. During the follow-up period of one year, the patient has been doing well.

## 3. Discussion

The larval form of *Echinococcus granulosus* is responsible for causation of more than 95% of human hydatid disease. *Echinococcus multilocularis*, on other hand, makes up for less than 5% of all cases (4). Hydatid disease is endemic to cattle rearing populations where the adult form of this parasite resides in the small intestine of the definitive hosts (dog) and is transmitted to intermediate cattle hosts (sheep) or accidental hosts (humans) by ingestion of embryo containing egg through contaminated vegetables/edibles. The embryo of the parasite passes via the portal vein to liver (first sieve) where majority of them are trapped forming cyst. If it escapes liver, the second

most common location is the lung (second sieve). If the embryo escapes both the sieves or by-passes them via lymphatics, it results in hematogenous and/or lymphatic dissemination, lodging the embryo in any part of the body to develop into a cyst (5). The larvae may also pass through the venous mesenteric lymph vessels by diffusion and get lodged in tissues and/or various intra-abdominal organs by transmural migration through the intestinal wall (6). If a microrupture occurs, direct spread from the adjacent sites may be another mechanism of infection (7). In our case, the transmural migration through the intestinal wall seems more plausible because neither hepatic nor pulmonary or any other adjacent organ was found to have been involved, in work up or during surgical inspection. Other unusual locations reported for cystic echinococcosis include spleen, kidney, peritoneum, retroperitoneum, mesocolon, omentum, pancreas, gallbladder, cerebral hemispheres, inguinal region, spinal cord, mediastinum, and seminal vesicle (8).

Hydatid disease in these atypical locations usually does not elicit symptoms except when it grows, produces local pressure symptoms or develops complications like rupture, secondary infection, or an allergic reaction (9). Retrorectal masses are a heterogeneous group of lesions which involve the space posterior to rectum and in front of sacrum. They can be both benign and malignant tumors and the majority of them are congenital and cystic in nature. Thus, differential diagnosis is difficult for the retrorectal masses and if hydatid cyst is suspected in the evaluation by immunological tests or imaging studies, unnecessary surgical procedures can be avoided. As evident from this case report and a few others, the diagnosis of a hydatid cyst should always be kept on the list as a rare possibility while assessing cystic swellings at any anatomical location in endemic areas.

At present, surgery is the most durable treatment modality in human cystic hydatid disease. The main purpose of surgery is to remove cyst without injuring the surrounding organs to which cyst maybe adherent. During the procedure, rupture of the cyst must be avoided at all costs to prevent recurrence. Alternatives for management of hydatid cyst including total cyst excision and partial cystectomy. Any approach employed should be thoroughly planned according to the location of the cyst and in relation to surrounding structures. In retro rectal lesions abdominal, perineal, or combined approach may be used depending on its position in relation to sacrum. In our patient, abdominal approach was used as the lesion was extending higher up in relation to the sacrum and simple enucleation was done to avoid damage to the rectal wall, urinary bladder, and pre-sacralplex uses, injury to which can have devastating consequences on patient's quality of life. Adjuvant Albendazole (10 mg/kg) treatment for a period of three-month was given to prevent postoperative recurrence.

#### 4. Conclusion

The diagnosis of hydatid cyst should always be kept in the bucket of differentials as a rare possibility while assessing

cystic swellings in retrorectal space in endemic areas of hydatid disease.

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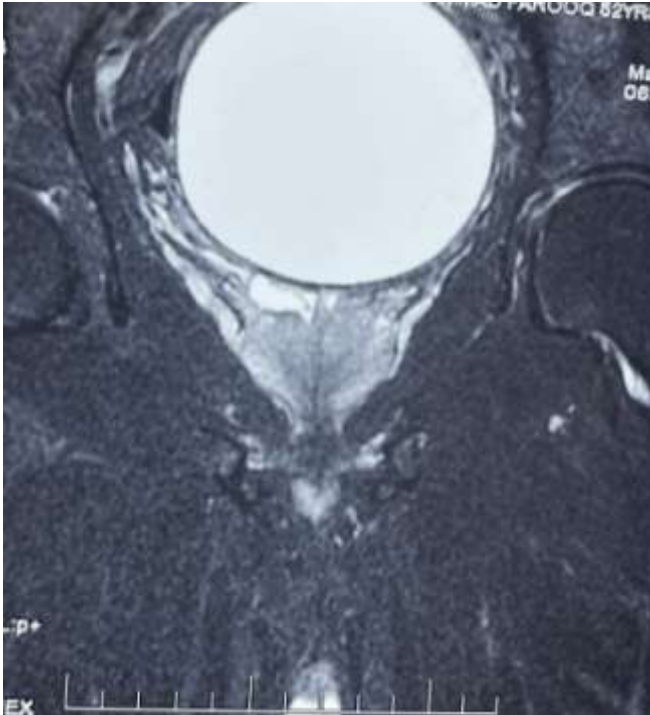


Image 1



Image 2

**Image 1 and 2:** MRI of pelvis coronal and sagittal cuts, T2WI showing cystic lesion between rectum and sacrum