

PELVIC HYDATID MIMICKING INGUINAL HERNIA

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PRESENTATION OF CASE

The classical finding in hydatid disease caused by *Echinococcus granulosus* with liver or lung involvement is well known. However, diagnosing hydatid disease at unusual locations maybe challenging even in endemic areas causing diagnostic confusion as primary extrahepatic extrapulmonary hydatid cysts are rare and only a few sporadic cases have been reported. We present a case where a painless progressive cystic swelling was present in left inguinal region with positive cough impulse and irreducibility mimicking an inguinal hernia. CECT was done, which confirmed it to be multiple hydatid cysts. The patient underwent excision of the cysts with mesh repair and left inguinal orchiectomy as the cord structures couldn't be separated from the cyst wall. Patient was discharged in satisfactory condition on oral antibiotics and albendazole. No recurrence noted on 6 months of follow up. So, a high suspicion for hydatid cyst should be kept in mind especially in endemic areas for any intra-abdominal swelling. The surgeon must keep in mind that this parasitosis can occur even in uncommon locations and therefore it should be regarded as a potentially systemic disease. In these cases, the diagnosis is challenging and can be achieved only through a complete interdisciplinary evaluation clinical, laboratory and radiological data.

DIFFERENTIAL DIAGNOSIS

There was diagnostic dilemma due to the location of the swelling in the inguinal region and the positive cough impulse and irreducibility on examination. A diagnosis of left incomplete irreducible inguinal hernia was made on clinical examination.

DISCUSSION

Hydatid disease is known since the ancient times and Hippocrates first described it as "liver filled with water." Famous Arab physician Al-Rhazes wrote about the disease more than one thousand years ago.¹ It is an infestation caused by the parasites of the order Cestoda, commonly known as flatworms and family taenia more commonly

known as tapeworms. Hydatid disease (echinococcosis) is a worldwide zoonosis produced by the larval stage of the *Echinococcus* tapeworm. The adult worms measure nearly 5 mm in length. The dogs, wolves and foxes are the primary hosts and cows, sheep, horses and pigs are the intermediate hosts. Humans act as accidental hosts.² Eggs of the organism are shed in the stools of the primary host. Human disease occurs when tapeworm ova are ingested by humans either by consuming unwashed and uncooked vegetables or as a result of close contact with a working or a pet dog.³ In humans, the two main types of hydatid disease are caused by *E. granulosus* and *E. multilocularis*. Although, the worldwide incidence and prevalence of Echinococcosis dramatically decreased in the last decades, it remains a major public health issue in several countries, where the infection is currently considered an endemic disease especially Mediterranean countries. These regions are all noted for the raising of sheep and cattle.² The State of Kashmir in India is endemic for hydatid disease. In endemic areas, any patient presenting with a cystic mass, in any tissue or organ, should be considered a potential case of the hydatid disease.⁴ It is important to be aware of the condition even in non-endemic parts of the world where only occasional cases are encountered, because of the rapid movement of large human groups from endemic to non-endemic areas.⁵

The tapeworm is composed of proglottid segments, which produce parasite eggs containing embryos (oncospheres). After being ingested orally, under the action of gastric and intestinal enzymes, the oncosphere is released; it penetrates the intestinal wall, joins the portal system and reaches the liver in the hepatic sinusoids.⁶ A few ova may pass through the liver (first filter) and reach the lung (second filter) and the systemic circulation causing hydatid disease in other organs. A possible dissemination through lymphatic channels has also been reported. This accounts for cases with solitary cysts in uncommon sites. The direct spread from adjacent sites maybe another mechanism of infection.⁷

The growth of hydatid cysts is usually slow and asymptomatic and clinical manifestations are caused by compression of the involved organ.² It depends upon the site of involvement, the size of the cyst and the pressure caused by the enlarged cyst. Usually, it presents as an inert, painless, non-inflammatory mass without any deterioration of the patient's general condition. However, if superinfected or cracked, the cyst can simulate an abscess or a cancer.⁷

The liver is the most frequently involved organ (75%), followed by the lung (15%). The diameter of the parasite is

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about 25 microns to 35 microns and it can pass through the hepatic sinusoids (10 microns to 100 microns) depending on the biophysical and structural features of the parasite, which has ameboid movements and different dimensions in the different stages of its biological cycle. It is as an emulsoid that adapts itself to the different diameters of the anatomic structures. Other factors such as pH, surface tension and the presence of the colloids are also involved in explaining the passage of the parasite through the hepatic sinusoids. However, it is also possible that the parasite reaches the peripheral locations by passing the hepatic sinusoids through the presence of arteriovenous anastomoses or through anastomoses between the portal and hepatic veins.⁷

Pelvic hydatid represents <2% in genitourinary area. Isolated hydatid cyst in extrahepatic and extrapulmonary sites often remain asymptomatic much like a slow growing benign tumour usually growing at the rate of 1-5 cm a year.² Cysts maybe single or multiple, uni- or multiloculated and thin- or thick walled.⁷

While the clinical signs of the hepatic cysts depend on its size and location inside the liver, when it is located in the peripheric organs, after an initial period during, which the cyst is asymptomatic, clinical signs appear only when the diameter of the cyst increases and determines "mechanical" clinical features, which will be different according to the organ where the cyst is located.⁷

Echinococcus multilocularis is more aggressive than E. granulosus and it may even mimic a malignancy.² The cyst of E. multilocularis differs from that of E. granulosus in that it grows by external budding of the germinal membrane with progressive infiltration of the surrounding tissue.⁸

The diagnosis of hydatid cyst relies on serologic tests and imaging techniques. Ultrasonography usually shows membranes, septations and daughter cysts within the cavity.² The sensitivity of various serological tests for hydatid disease varies from 64-87%. Imaging modalities remain more sensitive than serodiagnosis especially with unusual cyst locations and a characteristic CT scan demonstrating germinal membrane and daughter vesicles in the presence of negative serologic results should still suggest the diagnosis of echinococcosis.⁹ Enhancement of the pericystic soft tissues, mainly because of the collagen that is produced by the host can be considered an MRI feature suggestive of soft tissue hydatid disease.¹⁰ Cystic content is generally hypointense on T1 weighted images and homogeneously hyperintense on T2-weighted sequences. The endocyst may detach partially or completely from the pericyst causing "floating membranes" inside the cavity, a finding that is highly specific for hydatid disease. When there are daughter cysts inside the hydatid cyst, it appears multivesicular with a "honeycomb pattern."⁷

Notably, the higher rates observed in the elderly can be explained by the extensive incubation period of several years. Furthermore, no seasonality has been observed for Echinococcosis as expected for a disease with a very long incubation period.⁷

In cystic echinococcosis, the parasitic cyst consists of an inner germinal layer (endocyst) and an outer laminated layer

(ectocyst). The host reacts to the cyst by forming a fibrous capsule (pericyst), which contains blood vessels that provide nutrients for the parasite. From the germinal layer, scolices, brood capsules and daughter cysts are formed by endoproliferation (internal budding).¹¹



Figure 1. Showing Multiple Cysts in Left Inguinal Region

The main purpose of surgery is to prevent complications such as compression of surrounding structures, infection or cyst rupture.² During the surgical exploration after the identification of the cyst, it is surrounded by scolicedal solution impregnated gauze to prevent seeding of possible daughter cysts or other hydatid material. Uncomplicated giant cysts are mainly removed by needle aspiration. In that case, hydatid fluid is initially aspirated from the uppermost part of the cyst to decrease the intracystic pressure. A great caution is made when taking off the germinal membrane to avoid contaminating the neighbouring tissue. The residual cavity is sterilised by scolicedal solution (hydrogen-peroxide, povidone-iodine, hypertonic saline) according to the choices and habits of the surgeon. Postoperatively, patient can be discharged on oral albendazole as it interferes with glucose absorption through the wall of the parasite causing glycogen depletion and degenerative changes in echinococcal mitochondria.⁷

Diagnostic dilemma with hydatid cyst at unusual sites can lead to complications as sometimes it may present as acute surgical emergency or a chronic illness leading to morbidity.¹² Recurrent life-threatening anaphylactic shocks with or without laryngospasm maybe due to small, incomplete rupture of the cyst. Overt rupture of the cyst, however, is a much more frequent cause of anaphylaxis; it may be observed after trauma, even trivial, such as that derived from a sport (football) accident.⁷ Sometimes, bacterial super infection of hydatid cyst may occur. This results in pyogenic abscess formation within the cyst. Location of the cyst, type of organ, presence of intact pericyst and prone to trauma determines the risk of infection. Infection usually develops only after rupture of both the pericyst and endocyst.¹³

PATHOLOGICAL DISCUSSION

Histological examination confirmed multiple hydatid cysts.

DISCUSSION AND MANAGEMENT

A 61-year-old male admitted on 24/01/2015 with the complaints of swelling in left inguinal region for 3 months, which was insidious in onset, painless and progressive. There was no history of any altered bowel or bladder habits, weight loss, preceding trauma or fever. There was no history of exposure to pets. History of bilateral inguinal hernia repair 7 and 5 years back for right and left side, respectively. Patient was a known case of pemphigus vulgaris on regular treatment. On examination, scar marks on both the inguinal region was present of previous surgery. A cystic, nontender, irreducible swelling was present in left inguinal region with no cough impulse (Figure 1). The preoperative examinations (chest x-ray, full blood count, urine analysis and blood biochemistry) revealed no abnormalities. The hydatid serology was negative. Ultrasonography showed it to be 3 cystic swellings 5 x 3 cm, 3 x 3 cm and 4 x 3 cm in the inguinal region. CECT abdomen confirmed the USG findings (Figure 2). The patient underwent excision of hydatid cyst with meshplasty with left orchidectomy as the cord structures couldn't be separated from the cyst. Intraoperatively, there were multiple hydatid cysts with extension deep to sheath. The cavities were irrigated with 10% Betadine for 10 minutes and the defect was covered with Vicryl mesh. The spread of the hydatid fluid was prevented by putting around 4 to 5 surgery pads. No other abdominal cystic mass was found. A closed negative suction drain was put and skin closed. Starting from postoperative day 2, the patient was started on oral treatment with albendazole for 3 months. Patient was discharged on postop day 5. The patient has been followed for 6 months and no recurrence of hydatidosis has been detected.



Figure 2. CECT Showing Multiple Cysts in Left Inguinal Region

FINAL DIAGNOSIS

Hydatid cyst should be considered in the differential diagnosis of every abdominal intraparietal cystic mass, especially in regions where the disease is endemic. CT should be done in suspected cases to know the relationship to surrounding structures as well. The best treatment is the total excision of the cyst. Postoperatively, the parasiticide treatment with albendazole should be initiated.

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