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Case Report

An unusual case of disseminated hydatid cyst of liver, spleen and urinary bladder

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Abstract

Hydatid disease remains a prevalent health issue in regions where it is endemic, particularly in livestock-producing countries. When humans inadvertently swallow the ova, they get infested with *Echinococcus*. Infections in humans are typically contracted during childhood, and symptoms may appear five to twenty years later. The liver and lungs are frequently affected by the parasite, while the spleen and bladder are rarely affected. Only 4% of abdominal hydatid illnesses involve the spleen, and their incidence ranges from 0.5 to 4% globally. Urinary tract hydatid disease (HD) is extremely rare, making up approximately 2% to 4% of all cases. Both computed tomography (CT) and ultrasound are useful imaging techniques for identifying hydatid cysts. The lesion showed the characteristic "spoke-in-wheel" or "cyst within cyst" look. To avoid complications from cyst rupture following surgery, preoperative diagnosis is essential. Here, we discuss a 32-year-old man who had been experiencing progressive, non-radiating left hypochondrial pain for a year when he arrived at the emergency room. Following the required examination, a hydatid cyst of the bladder was identified, along with spleen and liver. The disease was managed by surgical evacuation, which was verified by histological analysis.

Keywords: Cyst within cyst appearance, Hydatid cysts, Livestock-producing countries, Spoke in wheel appearance.

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1. Introduction

The parasitic species *Echinococcus granulosus* (EG), or less frequently *Echinococcus multilocularis* (EM), *Echinococcus vogeli* (EV), and *Echinococcus oligarthus* (EO), are responsible for causing hydatid disease (HD), a form of zoonosis.¹ In areas where pastoral farming and animal husbandry are practiced, the prevalence of disease is high. Humans serve as intermediate hosts, unintentionally becoming infected by the fecal-oral route after consuming the parasite's eggs in food tainted by the feces of dogs or wild predators, which are the organism's definitive hosts.² Humans are the dead end for the parasites. Even though the majority of human instances are asymptomatic, they can lead to potentially fatal consequences such as anaphylactic shock. In addition to the liver (55% to 60%), the lungs (20% to 30%), kidney (2.5%), the heart (2.5%), bones (2%), muscle (1%), brain (0.5%), and spleen (1.5%), cysts can also be detected in

the omentum of the bladder, ovaries, parametrium, pelvis, thyroid, orbit, or retroperitoneum.^{3,4} Here we present a case of 32 year old male from rural areas of india who has concurrent involvement of urinary bladder, spleen and liver at the same time.

2. Case Report

The patient, a 32-year-old man, complained of increasing, non-radiating left hypochondrium pain that had been persistent for more than a year. Hepatosplenomegaly was evident during physical examination. The patient reported having a hepatic hydatid cyst that had been operated on in 2017. There was nothing noteworthy about his family history. Additionally, the patient's standard laboratory tests were completely unremarkable. Multiple hepatic and splenic cysts were discovered during a USG evaluation of the abdomen. As a result, a pelvic and abdominal CT scan was recommended. The CT scan showed a large well circumscribed

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peripherally enhancing cystic lesion measuring approximately 6.3x7.5x6.0 cm with multiple enhancing septa and few calcified foci involving segment II/IV of the left lobe of the liver. Another multiloculated cystic lesion of size 4.5x4.5 cm was noted in segment of right lobe of the liver. A multiloculated exophytic cystic lesion with honeycomb pattern of size 5.7x9.6x9.6 cm was seen arising from upper part of the spleen in sub diaphragmatic region. In the pelvic area next to the left lateral wall of the bladder, there was another sizable multiseptal lesion measuring 5x5.5x5 cm. A total splenectomy for a splenic hydatid cyst and a cystectomy for a liver and urinary bladder cyst were then planned for the patient (Figure 1-4).

Hydatid illness, were also visible in the cyst's inner membrane (Figure 6,7). Medical treatment was continued, and the patient was asked to come for followup.

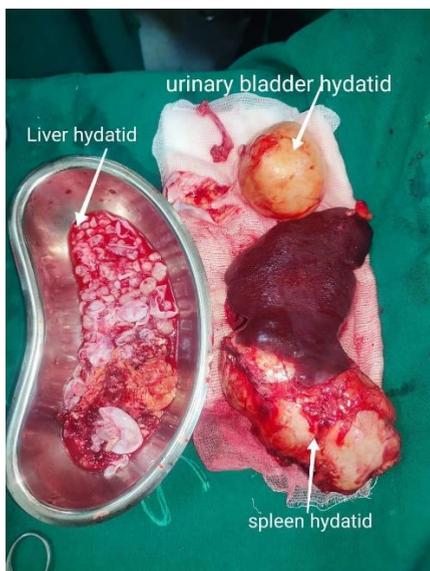


Figure 1: Cyst from liver and urinary bladder cyst and hydatid cyst with whole spleen



Figure 2: Spleen cyst

Pre and post-operative medical treatment was given. Upon gross examination, the specimen showed several white, smooth-surfaced cyst-wall fragments (Figure 5). The inner surface of the cyst-wall fragments revealed numerous tiny cystic structures attached to the larger cyst-wall that were filled with clear watery fluid. Under a microscope, the cyst wall's cross-sections showed an inner germinal membrane with a nucleated lining and an outer acellular laminated membrane. Numerous oval-shaped protoscolices with hooklets protruding from the membrane, indicative of



Figure 3: Liver cyst



Figure 4: Urinary bladder cyst



Figure 5: Numerous tiny cystic structures attached to the larger cyst-wall

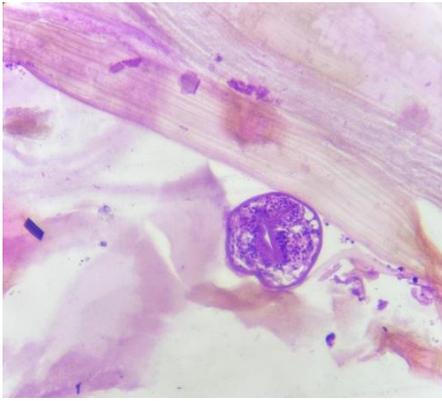


Figure 6: High power image shows brood capsule and scolices with visible hooklets Hematoxylin and Eosin (H&E)

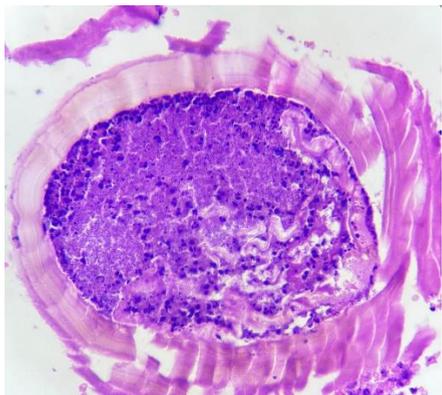


Figure 7: High power image of cyst shows acellular laminated layer Hematoxylin and Eosin (H&E)

3. Discussion

Echinococcosis has a more than two millennium-long history and is listed by the World Health Organization as one of the 20 neglected tropical illnesses.⁵ The zoonotic disease known as hydatid cyst is brought on by either adult or larval *Echinococcus granulosus* tapeworms. The life cycle of the parasite depends on interactions between two mammalian hosts. The intermediate host is a vegetarian, such as a sheep, whereas the definitive host, a carnivore, such as a dog, contains the tapeworm stage in its intestines. The eggs are excreted by the affected carnivores. Humans are the unintentional intermediary host,¹² who consume eggs that hatch in their intestines and release oncospheres, which usually settle and grow into larvae. After hatching from intestinal eggs, larvae can invade blood vessels and spread to almost every part of the body.⁶ The larvae typically enter the liver by the portal channel, but occasionally they manage to pass the liver barrier to reach the lungs and all other internal organs, where they develop into minute cysts.⁷

Hydatid cyst consist of three layers. The outermost is the fibrous tissue-based pericyst; the middle layer is the laminated, hyaline, and acellular membrane ectocyst; and the innermost is the endocyst, the germinating layer that produces daughter cysts and brood capsules with scolices asexually and secretes hydatid fluid internally and the

laminated membrane externally, generating new parasite generations. The cyst constitutes an inner germinal layer that asexually produces daughter cysts within the original cyst cavity.⁷⁻⁹ Lethal allergic reactions to parasite antigens may result from cyst rupture.^{6,7} The implantation of daughter cysts in other body organs, which leads to subsequent organ failure, is a more severe consequence of cyst rupture.^{6,7} The cysts may persist for a long time without producing any symptoms, and they often progressively grow to a diameter of 5–10 cm in a year.^{6,7} It typically manifests as abdominal pain and edema because it compresses the nearby organs and structures in a small area.

The parasites usually enter by the portal circulation, and most cyst embryos lodge in the liver and lungs. The parasite can also spread through the lymphatic system, retrograde migration from the vena cava to the subclavian vein, and, less frequently, the phenomenon of peritoneal fluid circulation.⁶ When the original cysts burst, either spontaneously or due to trauma, secondary cysts develop.⁹ Pelvic hydatid disease may be develop after retroperitoneal inoculation occur by penetration of rectum by larva and transmission to chest with lymphatic spread consequently.⁸

Computerized tomography (CT) scans combined with serological testing for the identification of echinococcal antigens in blood are the most effective diagnostic method.¹⁰ While ultrasound helps with diagnosis, a CT scan, which has a sensitivity of 90–100%, offers more detailed information.¹¹ There are five different types of USG findings in hydatid cysts: (1) simple cysts without internal architecture, (2) cysts with echogenic sand, (3) cysts with floating membranes (also known as "water Lily signs"), (4) calcified cyst and (5) multilocular cyst, which provides the impression of a "cyst within a cyst."¹² Though the presence of daughter cysts on CECT is pathognomonic, in some situations the final diagnosis is possible only on exploration. Numerous serological assays, such as immunoelectrophoresis, enzyme linked immunosorbent assay (ELISA), latex agglutination, and indirect haemagglutination (IHA), are used for diagnosis, screening, and postoperative follow-up for recurrence. However, these tests are often negative because the capsule separates the parasite from the host's immune system.⁴

Depending on the size and quantity of cysts, treatment options may include surgery, percutaneous therapy, and medications like albendazole.⁷ Surgery is the main treatment for hydatid cysts. However, medical treatment before and after surgery decreases the size and viability of the cysts, making surgery easier and lowering the chance of recurrence.¹³ Medical treatment can be used for solitary cysts, cysts with a modest adventitial reaction, and cysts that are small (7 mm in diameter). The drugs used to treat helminths are based on benzimidazoles. The cyst may also be managed via percutaneous aspiration, injection, and reaspiration (PAIR). A percutaneous puncture is performed under imaging guidance, some of the contents of the cyst are

aspirated, a scolicidal substance, such as hypertonic saline or 95% ethanol, is injected for approximately 15 minutes, and the contents are then aspirated again. PAIR is done on individuals who either refuse surgery or for whom surgery is not suitable for any reason.¹⁴⁻¹⁶ Total or partial splenectomy is the usual therapy for splenic hydatid cysts.¹⁷

4. Conclusion

Disseminated hydatid cyst disease, though rare but serious occurrence, presents complex diagnostic and management challenges. Prompt identification backed by radiological, clinical, and serological evaluations is crucial. The prognosis of the patient is greatly influenced by both medical and surgical interventions. Long-term postoperative monitoring, which is mostly based on computed tomography, ultrasonography, and serology, is crucial to identifying any recurrence. The main goals of preventive are to stop the parasite's life cycle, save cattle, and manage the dog population.

5. Source of Funding

None.

6. Conflict of Interest

None.

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