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Case Report

Primary Hydatid Cyst of Spleen: A rare entity

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Abstract

Primary hydatid disease of the spleen is very rare, even though spleen is the third most common organ involved in hydatid disease. Hydatid disease, a zoonosis, occurs worldwide but it is endemic in farming and cattle rearing areas. Most common sites of involvement by hydatid disease are liver followed by lung. Other rare sites include heart, pancreas, muscles and brain. The rarity of splenic hydatid disease poses a diagnostic challenge for clinicians especially in non endemic areas. We report a case of histologically confirmed primary hydatid cyst of spleen in a 23 years old female. This case report emphasises that hydatid disease should be considered in the differential diagnoses of all cystic masses in the spleen/ abdomen.

Keywords: Hydatidosis, Spleen, Echinococcus

1. Introduction

Hydatid disease (Echinococcosis) is a zoonotic infection caused by the larval form of parasites of tapeworm, Echinococcus granulosus. It is endemic in the sheep and cattle raising countries of Middle East, North Africa, New Zealand, Australia and South America[1]. The most commonly involved organ is the liver (75%), followed by lung (15.4%) and the spleen (5.1%)[2]. Hydatid disease is rarely encountered in the spleen, kidney, bone, thyroid, breast and pancreas[1].The worldwide incidence of splenic hydatid disease is 0.5- 4%[3]. In India, the recorded prevalence of the splenic hydatid cyst is 2.5% with highest incidence reported in central India[4]. The first case of splenic hydatid cyst was reported by Berlot in 1790 from an autopsy[5]. We report a rare case of primary splenic hydatid cyst in a 23 years old female from Haryana, India.

2. Case Report

A 23 years old female patient, farmer by occupation presented to the Surgery Outpatient Department with the complaint of dull aching pain in left hypochondrium for last 8 months. The patient also complained of malaise with nausea, vomiting, intermittent fever and weight loss since 6 months. She had no history of jaundice, cough, respiratory distress or abdominal trauma and her past medical history was unremarkable. On examination, her vital parameters were within normal limits. Physical examination revealed mild splenomegaly; mild tenderness in left hypochondrium was noted on palpation. Routine laboratory investigations viz. complete blood counts, coagulation profile, renal function tests, liver function tests & serum electrolytes revealed no abnormalities. ESR was 58mm/ 1sthr (Westergren). Abdominal Ultrasonography showed a round, well defined, cystic lesion of approximately 100x85mm over pancreas. Abdominal Computed Tomography (CT) scan revealed a large homogenous cystic lesion in spleen involving full length of splenic parenchyma measuring 10.5x8.5x8 cm with an imperceptible wall. All other abdominal and pelvic organs were unremarkable. Splenectomy with cyst removal was performed and the specimen was sent for histopathological examination. On gross examination, the spleen along with cyst measured 14x8x8cm and weighed 820gm (Fig. 1). On cutting open, 650ml of hydatid sand and fluid came out. The outer surface of cyst was congested. The inner surface was glistening grey white.

Fig.1: Specimen of spleen along with cyst.

Histopathological examination showed scattered brood capsules with hooklets along with the cyst wall having an outer acellular laminated layer and inner germinal layer characteristic of hydatid cyst (Fig. 2a, 2b, 2c & 2d).

Fig 2: Histopathological examination

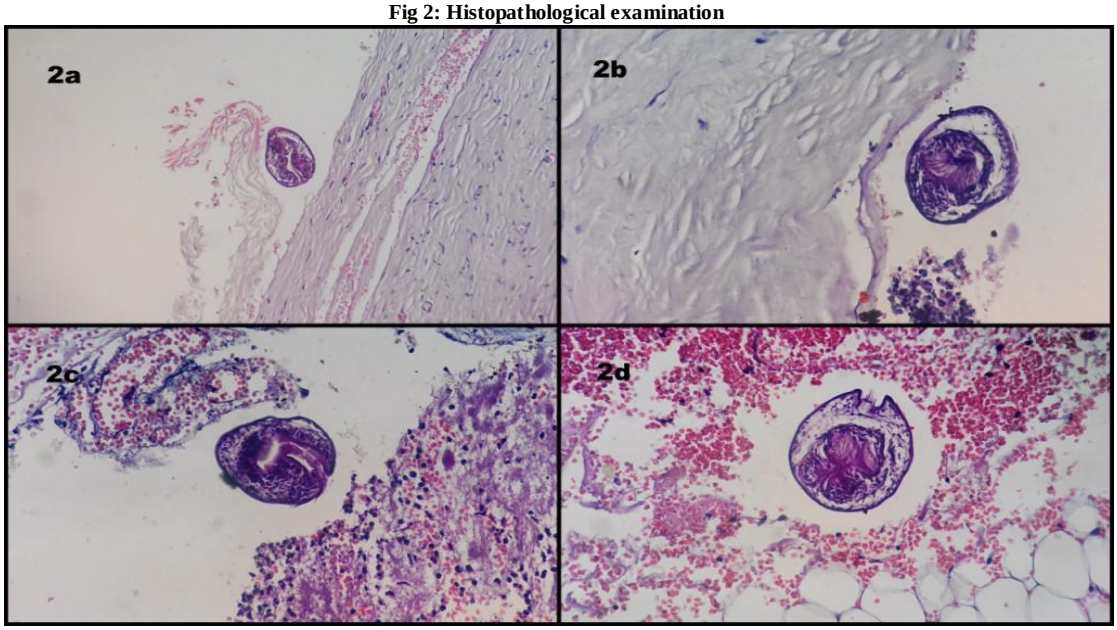

Fig. 2a: Microphotograph showing scattered brood capsule along with an outer acellular laminated layer (H&E; x100).
Fig. 2b: Microphotograph showing scattered brood capsule with hooklets along with an outer acellular laminated layer (H&E; x400).
Fig. 2c&2d: Microphotograph showing scattered brood capsule with hooklets along with congested splenic tissue (H&E; x400).

A final diagnosis of splenic hydatid cyst was made. The patient was vaccinated prior to splenectomy with Hib and Meningococcus C conjugate vaccines to decrease the risk of overwhelming post splenectomy sepsis (OPSS) and splenectomy was performed thereafter. Patient was given preoperative Antihelminthic drugs. Before discharge, patient was educated about the risks of OPSS and was advised to continue Antihelminthic treatment.

3. Discussion

Parasitic cysts of the spleen are almost exclusively hydatid cysts. In endemic areas, 50-80% of splenic cysts are echinococcal[2]. Four species of Echinococcus cause infection in humans; E. granulosus and E. multilocularis are the most common, causing cystic echinococcosis and alveolar echinococcosis respectively. The two other species; *E. vogeli* and *E. oligarthrus* cause polycystic echinococcosis and are less frequently associated with human infection[2]. The life cycle of Echinococcus includes a definitive host (usually dogs or related species) and an intermediate host (such as sheep, goats, or swine). Humans are incidental hosts; they do not play a role in the transmission cycle[2].

After ingestion, the eggs hatch and the resultant oncospheres penetrate the intestinal mucosa of humans and enter the circulation. They primarily lodge either in the liver, lungs or kidneys which act as filters. In the organs where these organisms lodge, they slowly develop a cavity lined with germinal epithelium, outside which develops a laminated acellular area[6].

Primary infestation of the spleen usually takes place by the arterial route after the parasite has passed the two filters (hepatic and pulmonary). A retrograde venous route through portal circulation which bypasses the lung and liver is also reported. Secondary splenic hydatid disease usually follows systemic dissemination or intraperitoneal spread following ruptured hepatic hydatid cyst[7].

Hydatid cysts are usually slow growing and in some organs, especially in the spleen, they can reach an enormous size and be asymptomatic and incidentally diagnosed[2]. The main symptoms associated with the disease are abdominal discomfort, pain and palpable mass in left hypochondrium[1]. Our patient presented with these signs and symptoms.

The complications of untreated splenic hydatid cyst are mainly secondary infection, inflammation, anaphylactic shock, intraabdominal rupture of cyst, acute abdomen, and compression of other viscera or fistulisation to the bowel, mainly colon. Pericystic inflammation may cause adhesions with nearby organs such as the kidney, left diaphragm, colon, stomach[1].

The main problem in the diagnosis of splenic hydatosis is in differentiating it from other splenic cystic lesions that have similar appearances on sonography and computed tomography (CT). The differential diagnosis of such lesions includes epidermoid cyst, pseudocyst, large solitary abscess or hematoma, intrasplenic pancreatic pseudocyst and cystic neoplasms of the spleen[8].

At present sonography and CT are the most valuable imaging techniques in the diagnosis and evaluation of focal splenic diseases[8]. CT has higher overall sensitivity than ultrasound, with sensitivity rates of 95-100%. Plain radiograph may demonstrate a marginal/ crumpled egg shell like calcification in the splenic area suggestive of splenic hydatosis. Serological tests such as ELISA, immunoelectrophoresis or indirect hemagglutination test, when combined with imaging, can lead to the successful diagnosis of splenic hydatid disease in 90% of the cases.[2] In our case, imaging studies were suggestive of splenic hydatid disease but the diagnosis was confirmed only after histopathological examination. On histopathology, hydatid cyst consists of three layers. The outer most adventitia is formed of compressed splenic tissue, the middle layer, laminated membrane of friable ectocyst and an innermost germinal layer, endocyst from which large number of scolices are produced.[1]

Total splenectomy, partial splenectomy, cyst enucleation and unroofing with omentoplasty are the various preferred surgical techniques to treat splenic hydatid disease.[9] Many trials are usually made for conserving the spleen, so as to avoid overwhelming post splenectomy sepsis (OPSS). Partial splenectomy carries a risk of poor vascular control when incising the splenic tissue while unroofing the cyst wall leaving behind a residual cavity carries the risk of post operative infection. For the above reasons and the possibility of multiple splenic cysts, total splenectomy should be the method of choice.[1] However during surgical treatment extreme caution must be taken to avoid life threatening complications like anaphylactic shock due to spillage of cyst contents. Laproscopic approach has also been advocated for uncomplicated hydatid cyst of the spleen. Chemotherapy and newer methods, such as puncture aspiration, injection and reaspiration (PAIR)

technique using hypertonic saline or 0.5% silver nitrate solution before opening the cavities tend to kill the daughter cysts.[1] Our patient was treated with total splenectomy. Antihelminthic drug therapy (Albendazole and praziquantel) were given in pre and post operative period. Prior to splenectomy, the patient was given prophylactic vaccination against *Streptococcus pneumoniae*, *Hemophilus influenza* type b and *Neisseria meningitidis*, and was started on six month course of prophylactic penicillin. No post splenectomy infection was encountered.

4. Conclusion

Hydatid cyst should be considered in the differential diagnosis of all cystic masses in the spleen/ abdomen, especially in endemic areas. An early diagnosis and careful preoperative evaluation is recommended to avoid further complications.

Conflict of Interest

There is no conflict of interest.

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