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Case Report

Splenic hydatid cyst: a case report

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ABSTRACT

Hydatid cyst (HD) disease is a parasitic infection caused by *Echinococcus granulosus*. It occurs worldwide, but its prevalence is higher in the rural communities where sheep and cattle raising constitute a crucial component of the agricultural industry. This is a zoonotic disease which spreads via close contact of humans with sheep and dogs. The most common organs involved in hydatid cyst are the liver and lungs. However, other organs may also foster it, such as the spleen which is a rare clinical condition, with an incidence rate of between 0.5% and 4%. In this study, a 46-year-old male with a primary splenic HC is reported with chief complaint of pain and non-tender palpable mass in left hypochondrium. A 9.7×10.6×11 cm splenic cystic mass was detected on ultrasonography (US) and confirmed by abdominal computed tomography (CT) scan, without involvement of other organs. Splenectomy was done along with excision of intact hydatid cyst. Pathological examinations revealed cystic hydatidosis. We describe this case of an isolated splenic HC, which was successfully treated with total splenectomy, focusing on the management and outcome of this disease.

Keywords: Echinococcus granulosus, Zoonotic disease, Spleenic hydatid cyst, Spleenectomy

INTRODUCTION

The involvement of spleen in hydatid disease (HD) is rare. Cases of splenic hydatid cyst (SHC) can be either primary (isolated to the spleen only) or secondary (accompanied by hydatid cysts in other organs). Primary splenic involvement by hydatid cyst is rare and accounts for less than 2% of patients.[1] Berlot was first to introduce Primary splenic hydatidosis in 1790.[2] Splenic involvement is uncommon event because cyst embryos are trapped in the liver and lungs, with only 15% entering systemic circulation. The parasite's eggs exit the liver-lung barrier, initiating primary infestation of the spleen through the arterial route. Splenic hydatid disease can also occur through retrograde parasite spread via the portal and splenic veins, bypassing the lung and liver.[3] Secondary splenic hydatid disease typically results from systemic dissemination or intraperitoneal spread after the rupture of a hepatic hydatid cyst. The main presentation of splenic hydatid disease is a slowly growing, asymptomatic cystic mass, although it may manifest as a painful bulge in the left hypochondrium. Dyspnea and anaphylactic responses may occur due to cyst rupture. The atypical location of hydatid cysts in the spleen can complicate the differential diagnosis, as they may appear as solitary or multiple multiloculated cysts.[4]

CASE REPORT

A 46-year-old male came to the hospital with chief complaints of pain abdomen in upper left quadrant. The patient developed pain in abdomen, which was gradual in onset, diffuse and progressive in nature. It was associated with intermittent nausea.

There was no history of fever, vomiting, jaundice, bowel and bladder complaints. No history of known comorbidities. No significant past history.

On general examination, pulse rate (PR): 82/min, blood pressure (BP): 130/80 mmHg SpO_2: 99% afebrile. Per abdomen examination: soft, nondistended, palpable mass in left hypochondrium, non-tender, bowel sounds present.

Routine investigations, haemoglobin (Hb): 13.4 g%, total leucocyte count: 7.81 mg/dl eosinophils: 12%, platelet count: 227000/cumm, total bilirubin: 0.24 mg/dl, direct bilirubin 0.08 mg/dl, serum glutamic oxaloacetic transaminase 16 U/l, serum glutamic pyruvic transaminase 16 U/l, alkaline phosphatase 111 U/l, T. protein: 6.4 g/dl, albumin: 3.7, urea: 21 mg/dl, creatinine: 0.74 mg/dl, S. sodium: 141 meq/l, potassium: 4.5 meq/l and chloride: 104 meq/l. On radiological investigations, USG (W/A) showed multisepatated cystic lesion in splenic parenchyma hydatid cyst. Contrast enhanced computed tomography (CECT) whole abdomen showed a large multiseptated cystic lesion in left hypochondrium measuring approximately 9.7×10.6 cm in cross section and approximately 11 cm in craniocaudal extent the lesion has with suspicion of few peripherally arranged cysts in it closely abutting the posterior wall of the stomach and tail of pancreas. Exophytic splenic lesion - s/o splenic hydatid cyst.

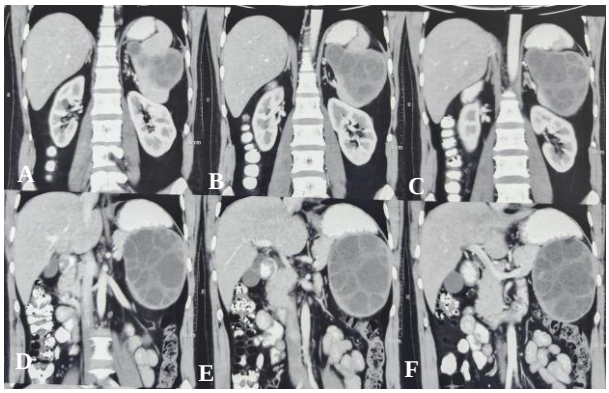

Figure 1 (A-F): CECT coronal section showing spleen with multiloculated hydatid cyst.

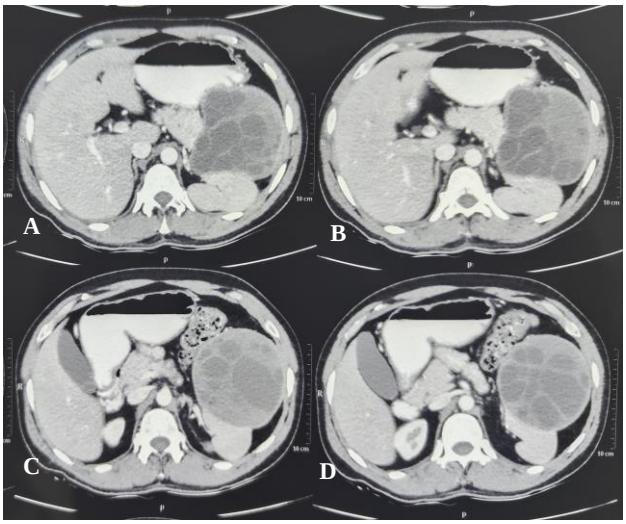

Figure 2 (A-D): Transverse section showing spleen with multiseptated cystic lesion s/o hydatid cyst.

Patient was planned for laparoscopic pericyctectomy under GA but due to large size of hydatid cyst and more than 80% involement of spleen, the procedure was converted to open splenectomy. Intra-operative findings revealed spleen with hydatid cyst measuring 11×10 cm.

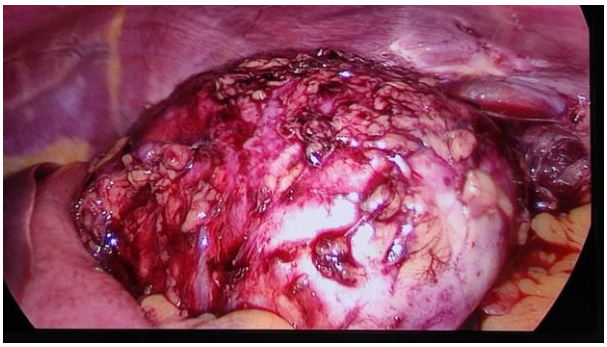

Figure 3: Intra-operative picture showing intact splenic hydatid cyst.

Splenic vein and artery were identified and ligated and cut. Spleen and hydatid cyst was separated completely from the surrounding tissues extracted out en masse. Abdominal drain was placed and Abdomen closed in layers. Post-operative stay was uneventful.

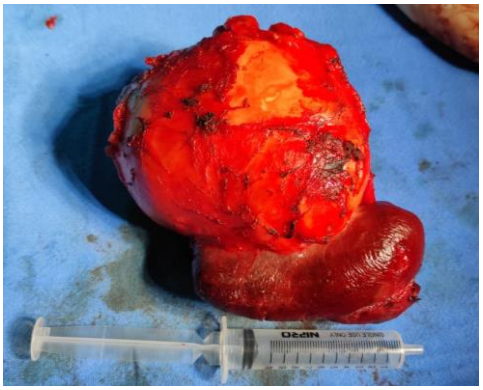

Figure 4: Resected spleen with hydatid cyst measuring approximately 10×10 cm.

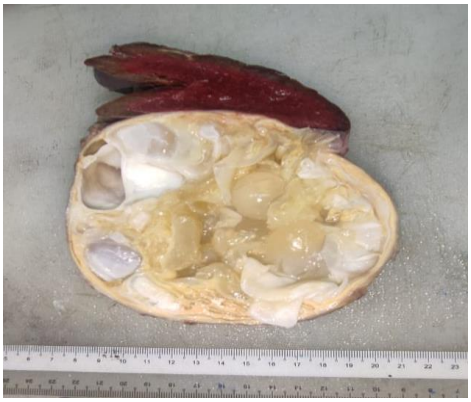

Figure 5: Gross splenectomy specimen measuring 11×9×8 cm. The hilum shows a large hydatid cyst measuring 11×5×4 cm showing daughter cysts.

Histopathology report

Received a splenectomy specimen measuring 11×9×8 cm. The hilum shows a large cyst measuring 11×5×4 cm, filled with clear serous fluid. Multiple daughter cysts are also seen within the large cyst comprising of collapsed whitish membranous structure with yellowish granular material adherent to the wall. Grossly cyst not appearing to invade the splenic tissue. Microscopically section show normal splenic tissue, seen attached to the capsular surface is a cyst with wall composed fibrocollagenous host tissue infilterated by chronic inflammation admixed with eosinophils (pericyst). The inner layer is composed of pale, eosinophilic laminated chitinous layer (ectocyst). The inner germinal layer shows markedly degenerated eosinophilic material with few showing proto scolices and hooklets embedded within them.

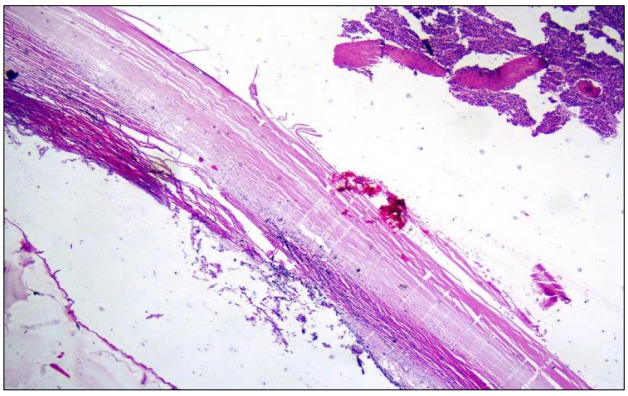

Figure 6: Low power photomicrograph from the splenic cyst shows splenic tissue (short arrow), pericyst (long arrow) and laminated membranes (arrow head) (H and E, at 40X).

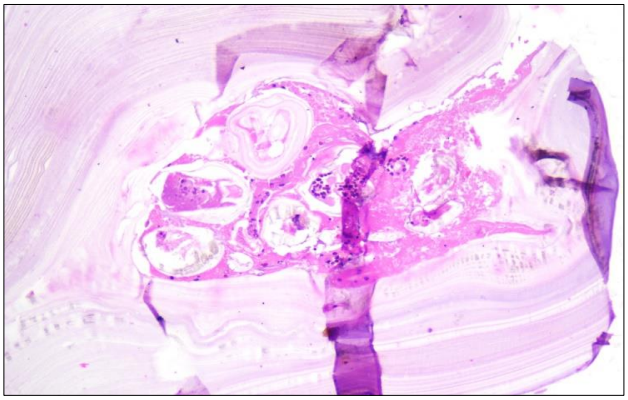

Figure 7: Low power photomicrograph from the splenic cyst shows splenic tissue (short arrow), pericyst (long arrow) and laminated membranes (arrow head) (H and E, at 40X).

DISCUSSION

Hydatid disease poses a significant global health concern, particularly prevalent in regions where sheep and cattle farming is widespread. The most common sites of hydatid disease are the liver (60-70%), which acts as a first filter and the lungs (10-40%), which acts as second filter. Uncommon locations for hydatid cysts encompass the spleen, thyroid, gallbladder, central nervous system, kidney, psoas sheet, retroperitoneal region, and orbit. The hydatid cyst is structured with three layers: the outermost adventitia (pseudo cyst) consists of compressed splenic tissue, a middle layer constitutes a laminated membrane of fragile ectocyst, and the innermost layer is the germinal layer, endocyst. Splenic hydatid cysts typically exhibit asymptomatic, solitary, slow growth and are often incidentally discovered. Common symptoms include abdominal discomfort, pain, and the presence of a palpable mass in the left upper quadrant. The complications of untreated splenic hydatid cyst are mainly secondary infection, and acute abdomen, compression of other viscera,spleen atrophy, intraabdominal rupture and fistulization to the bowe.

Teke et al reported a splenic hydatid cyst perforating into the left colon can result in significant gastrointestinal bleeding.[8] There have been documented cases of ruptured splenic hydatid cysts leading to a spleno-thoracic fistula, causing communication with the thoracic cavity. Instances of severe anaphylactic reactions following cyst rupture are also reported, characterized by fever, pruritus, dyspnea, stridor, and facial edema. Differential diagnoses for splenic hydatidosis include cystic spleen lesions like abscesses, epidermoid cysts, hematomas, post-traumatic pseudocysts, and neoplasms such as lymphangioma and hemangioma. The pre-operative diagnosis can be challenging due to similarities in presenting symptoms and radiological findings with more commonly encountered splenic lesions. Eosinophilia may be the finding on haematological investigation.

The Casoni skin test is sensitive but not specific. Radiological diagnosis through plain X-ray, ultrasonography (USG), CT, and MRI serves as a valuable method for identifying hydatidosis. In abdominal or chest radiographs, the presence of marginal or crumpled eggshell-like calcifications in the splenic region indicates potential splenic hydatidosis. Ultrasonography and computed tomography are primary diagnostic tools for detecting splenic hydatid cysts. Serological tests exhibit high sensitivity and specificity for Echinococcosis. Despite the limited effectiveness of drug therapy and the risks associated with spontaneous or traumatic rupture, the surgical approach remains the established standard for managing hydatid disease. The standard treatment is splenectomy as Complete resection removes all parasitic and pericystic tissues.[5]

In surgical interventions, it is crucial to exercise utmost care to prevent cyst rupture. The preferred surgical approaches for treating splenic hydatid disease include total splenectomy, partial splenectomy, cyst enucleation, and unroofing with omentoplasty. Many trials are usually made for conserving the spleen, so as to avoid

overwhelming post splenectomy sepsis (OPSI).[6] 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 postoperative infection.[7] Given the potential for multiple splenic cysts and the risk of cyst rupture, total splenectomy is the preferred approach, especially when there is communication between the spleen and neighboring organs like the stomach, colon, and diaphragm. The laparoscopic method is also recommended for uncomplicated hydatid cysts of the spleen. Chemotherapy and innovative techniques, such as the puncture, aspiration, injection, and re-aspiration (PAIR) method using hypertonic saline or 0.5% silver nitrate solutions before opening the cavities, aim to eliminate daughter cysts. Safe percutaneous drainage of uncomplicated hepatic hydatid cysts leads to cyst disappearance, offering advantages like a shorter hospital stay and a reduced complication rate.[9]

Post-operatively, medical treatment becomes crucial. Antihelminthic drug therapy involves the use of Benzimidazole chemotherapy drugs such as albendazole (10-15 mg/kg/day for one month) or mebendazole (40-50 mg/kg/day for 3-6 months). Additionally, praziquantel (40 mg/kg/week for 2 weeks pre and postoperative) is administered to minimize the risk of anaphylactic shock and decrease tension in the cyst wall. Albendazole proves effective as an adjuvant therapy in hydatid cyst treatment, significantly reducing the likelihood of recurrence in patients who receive this therapy. Gil-Grande et al reported that albendazole effectively renders 72.3% of cysts sterile within the initial month, with a further increase to 94% by the conclusion of a three-month treatment period.[10] In our specific case, a laparoscopy-assisted total splenectomy was performed due to spleen involvement exceeding 80%. Following the splenectomy, the patient received preventive vaccinations against *Streptococcus pneumoniae*, Haemophilus influenza type b, and *Neisseria meningitidis*. Additionally, a six-month regimen of prophylactic penicillin and benzimidazole chemotherapy was initiated. No post-splenectomy infections were observed.

CONCLUSION

The rarity of occurrences poses a significant early diagnostic challenge for splenic hydatid disease, particularly in non-endemic regions. Its common presentation involves a nonspecific painful abdominal mass. The splenic hydatid cyst may become a challenging surgical problem. Preoperative evaluation should be carried out carefully. Computed tomography scan is the most sensitive investigation for diagnosis. Although the management must be individualized for each patient, a surgical resection is the best curative procedure. Postsurgical pharmacological treatment is necessary to ensure complete healing.

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