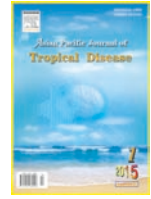




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Isolated large primary splenic hydatid cyst: A case report

Manish Garg^{1*}, Abha Mangal², Harsh Tak³, Dharmendra Pal Singh⁴, Atul Soni⁴

¹Department of General Surgery, Max Healthcare Pvt. Ltd. Gurgaon, Haryana, India

²Department of Community Medicine, Vardhman Mahavir Medical College and Safdarjung Hospital, New Delhi, India

³Department of General Medicine, JLN Medical College, Ajmer, Rajasthan, India

⁴Department of General Surgery, ESI Model Hospital, Gurgaon, Haryana, India

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ABSTRACT

Hydatid cysts of the spleen are uncommon (2-3.5%). Primary involvement of the spleen is even rarer. We present a case of a large primary hydatid cyst of the spleen in a 55-year-old woman. Diagnosis was based on computed tomography (CT) scan along with a high degree of clinical suspicion. En-masse excision was successfully performed, and the patient was discharged with prophylactic albendazole therapy. Unless there is a high degree of clinical suspicion, this life-threatening condition may be missed; hence, the importance of this report.

1. Introduction

Hydatid disease, or human echinococcosis, is a zoonotic disease caused by the larval forms (metacestodes) of the tapeworm, *Echinococcus granulosus*[1-3]. Dogs and other carnivores are its definitive hosts, while the most common intermediate hosts are sheep and cattle; humans are incidental hosts. Infestation occurs through fecal-oral route on ingestion of food contaminated with the feces of infected animals[2,3]. The prevalence of infestation among food animals in India is reported to be high[1]. A study from Delhi reported that 10% of the slaughtered sheep are infested[4].

The liver is the most common site of infestation in the majority of patients (60-75%) since it acts as first filter[1,2]. The second most common site is the lung (10-25%). Other organs, such as the spleen,

kidneys, heart, bones, brain, etc., are less frequently involved[1,2,5]. Among abdominal hydatid cysts, splenic hydatid cysts are quite rare (2-3.5%)[2,3,5]. Primary and isolated involvement of the spleen is even more rare[5,6]. It may be detected incidentally or present with nonspecific complaints, and should be suspected in all cystic lesions of the spleen in endemic areas[2]. Prompt treatment is mandated as traumatic or spontaneous rupture may lead to life-threatening anaphylaxis[3].

2. Case report

A 55-year-old female from a rural area in Haryana (India), working as a farmer in her own field presented to the surgery outpatient department with abdominal pain of two years' duration and a lump on the left side of the abdomen, which was gradually increasing in size over the previous three months. Physical examination revealed an asymmetric abdomen and a large lobular mass in the left upper abdominal quadrant. Routine laboratory investigations, liver and kidney

*Corresponding author: Dr. Manish Garg, Senior Resident, Department of General Surgery, Max Healthcare Pvt. Ltd. Gurgaon, Haryana, India.

Tel: +91 9871343114

E-mail: manishgarg11@gmail.com

function tests were normal. Ultrasound revealed a large cystic lesion measuring approximately 148 × 122 mm in the left suprarenal region, with multiple solid-appearing cysts located towards the periphery with central solid looking appearance. The spleen appeared normal with a calcified lesion measuring 22.4 mm. Other solid viscera appeared normal. CT scan revealed two calcified cystic lesions measuring 45 × 30 mm and 21 × 18 mm at the peripheral region of the spleen (Figure 1). Daughter cysts were visualized within these lesions. The normal splenic tissue appeared compressed due to the cyst. Other solid viscera were normal in appearance. There was a large calcified hydatid cyst occupying the left side of abdomen extending from the suprarenal region to the iliac fossa in the CECT findings. Immunological tests like ELISA and Immunoelectrophoresis were not done as it was not available in our setting. Immediately after admission, prophylactic therapy with albendazole (15mg/kg/day) was started in preparation for surgery. Patient was kept nil by mouth (NBM) for 6 hours prior to surgery.

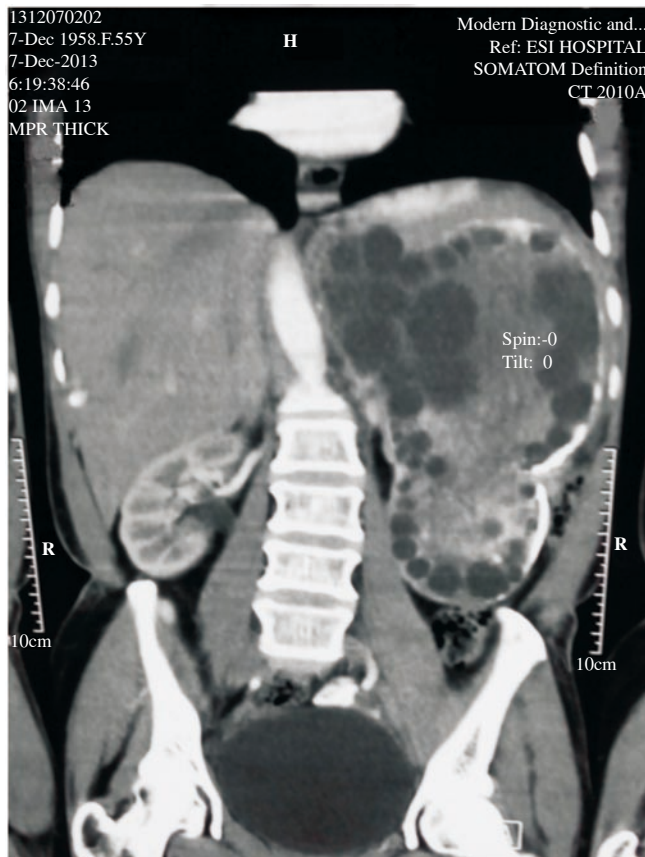


Figure 1. CT abdomen showing enlarged spleen due to hydatid cyst with multiple daughter cysts.

Exploratory laparotomy, on day 2 post-admission, revealed a large mass filling the entire left side of the abdomen, pushing the intestines medially. The cyst extended from the diaphragm to the left iliac fossa. Upon careful dissection, the spleen and hydatid cyst were removed en-masse (Figure 2). The postoperative period was uneventful and the patient was discharged on post-operative day-8 with albendazole therapy for 3 weeks. Follow-up visits (bimonthly) were uneventful. Patient remained asymptomatic at all follow-up visits and per abdomen

examination was normal so no imaging studies were done as she was a poor patient. At the two week follow-up visit of the patient she was given the vaccines for Hib, pneumococcus and meningococcus. Histopathological examination of the sample confirmed hydatidosis.



Figure 2: Gross specimen of spleen resected en-masse.

3. Discussion

Hydatid cysts are a common problem in farming communities[3]. Hydatid disease of the spleen is a rare condition even in endemic areas[1,2,3,5] as the liver, which acts as the first filter, is the most common site and the lungs, being the second filter, are the second most common site[1,2,3,5]. Only 15% of cyst embryos escape these two filters and enter systemic circulation, primarily via the arterial route, to affect other organs such as the spleen, which is the third common site of infestation[3]. Another route of rare primary splenic involvement is retrograde spread of parasites via the portal and splenic veins bypassing the liver and lungs[3]. Secondary involvement of spleen is more common than primary involvement following the systemic dissemination or intraperitoneal spread of ruptured hepatic hydatid cyst[3]. Splenic hydatid cysts are usually asymptomatic, solitary, and slow growing (rate of increase in size is 2-3 cm/year). They are often diagnosed incidentally[3]. The most common symptoms are abdominal pain and a mass in the left upper abdomen[3,7]. Previous studies have reported that pain in the abdomen is the most common complaint[3,8]. The differential diagnosis of splenic hydatid cysts includes other cystic lesions of the spleen, *e.g.*, splenic abscess, dermoid cysts, hematomas, pseudocysts, and neoplasms, such as lymphangioma and hemangioma[3,6,7,9]. The diagnosis of hydatidosis should be favoured in patients with splenic cystic lesions, especially in endemic area until proven otherwise[3].

Preoperative diagnosis is often difficult due to similarity in presenting and radiological features[3]. Serological tests like ELISA, immunoelectrophoresis (90-95% sensitivity) and indirect hemagglutination test (85% sensitivity) when combined with imaging studies can lead to successful diagnosis of splenic hydatid disease in about 90% cases[3]. Ultrasonography and CT scan are the major

diagnostic tools[3]. The sonographic findings of splenic hydatid cysts are not specific, but it is useful in the early stages, when the lesion is cystic, in detecting daughter cysts, hydatid membrane and hydatid sand[3]. It has a sensitivity of about 90-95%[3]. The most common appearance on sonography is an anechoic smooth and round cyst which is similar to that of a benign cyst[3]. Mixed echoes due to membranes are similar to the picture of an abscess or neoplasm[3]. Snake or serpent sign, spin/whirl sign, double line sign and the waterlily sign are classical signs on ultrasonography which when present help to differentiate hydatid cysts from other similar pathologies[3].

CT has a higher sensitivity than ultrasound, having sensitivity rate of 95-100%. It is the best modality for determining the number, size, and anatomic location of the cysts. It is more sensitive to detect subtle cyst wall calcification and extra hepatic cysts[3]. CT attenuation depends on the intracystic content. Hydatid cysts having homogenous fluid content show water attenuation values, while presence of intracystic debris, hydatid sand and inflammatory cells shows high CT values on unenhanced scans[3]. MRI is emerging as an important imaging technique for diagnosis and evaluation of hydatid disease. It is useful in patients with negative serology and indeterminate ultrasound and CT findings[3].

Due to limited efficacy of drug therapy and risk of spontaneous/traumatic rupture of cysts leading to life threatening anaphylaxis, surgery (open or laproscopic) is the standard approach for treatment[3]. Complete en-bloc resection of the spleen is the gold standard of treatment as it ensures the removal of all parasitic and peri-cystic tissues[3,6,7]. Splenectomy has a low morbidity and mortality rate and is the preferred treatment for large cysts where splenic parenchyma is significantly reduced due to pressure atrophy and the fibrous membrane is thin and fragile increasing the risk of intraoperative rupture of cyst[3]. Therefore, splenectomy must be carried out in large and giant hydatids of the spleen localized in the organ or it's hilum and in irreversible derangement of organ function[3]. Complications associated with splenectomy are hemorrhage, pancreatitis, gastric injuries and most importantly overwhelming post-splenectomy infection (OPSI). Therefore all such patients should be vaccinated against *Streptococcus pneumoniae*, *Haemophilus influenzae* type B, and *Neisseria meningitidis* to decrease the risk of OPSI. The Hib and meningococcus C conjugate vaccines should be given either at least two weeks before or two weeks post splenectomy[3].

Though surgery offers the possibility of immediate and total cure, it is not without risks. Secondary echinococcosis may occur in 2-21% cases due to spillage. 0.5-4% operative mortality has been reported[3]. Also, chances of recurrence are there if other small cysts are present but not resected, specially if a conservative approach is used. Reliable data on incidence of recurrence is not available, though it is estimated to be between 4.6% to 22.0% for hydatid disease and most often with more conservative procedures. It is never seen following complete resection of

an intact cyst[3].

Some studies recommend laparoscopic resection of spleen for uncomplicated hydatid cysts. If used for small-sized and superficially located cysts the success rate is similar to conventional surgery with lesser complications[3]. But if there are multiple cysts, large in size, located deep in the organ, or infected for the second time open surgery remains the standard treatment. Spleen preserving surgery may be considered in children to prevent OPSI depending on the merits of the individual case[3].

World Health Organization (WHO) recommends pre-operative prophylaxis with albendazole and praziquantal, started one month and two weeks prior to surgery respectively or at least four days prior to the surgery. It helps to stabilize cysts, decreases internal tension and reduces the incidence of anaphylaxis and recurrence[3]. Earlier it was an accepted practice to give albendazole (10-15 mg/kg/day) in the postoperative period for 28 days to decrease recurrence[3,7]. But no long term studies are available for evaluation of it's efficacy in prevention of recurrence as well as it's optimal schedule. There is a need for accurate guidelines in this regard[3].

Follow-up for at least three years post surgery is mandatory to determine the efficacy of treatment in cases where conservative or spleen preserving approach has been used. It is best done by ultra sonography[3].

Conflict of interest statement

We declare that we have no conflict of interest.

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