



GIANT SOLITARY SPLENIC HYDATID CYST: A CASE REPORT AND REVIEW OF LITERATURE.

Surgery

**Dr. Mohit M
Agrawal**

Consultant Surgeon Sanjeevani Multispeciality Hospital, Mumbai

**Dr. Tejinder Singh
Chhabda***

Associate Prof., Dept. Of Surgery MGM Medical College, Aurangabad *Corresponding Author

ABSTRACT

Primary hydatid disease of the spleen is a very rare condition. Usually splenic hydatid cysts are secondary, resulting from either spontaneous spread of cysts or occurring after surgeries involving hydatid cysts in other regions. Even in the endemic region, the frequency is reported to be 0.5 – 4% of abdominal hydatid diseases [8]. The most common sites of hydatid disease are the liver (60–70%), acting as a first filter followed by the lungs (10–40%), which acts as second filter. The other rare sites include spleen, kidney, thyroid, gall bladder, central nervous system and retroperitoneal region.

We report a case of a primary isolated hydatid cyst of spleen treated by a classical surgical approach. This case report and literature review describes the management of splenic hydatidosis.

KEYWORDS

Hydatid cyst; spleen; hydatidosis; Echinococcosis

Introduction

Hydatid disease or Echinococcosis is a zoonotic infection caused by the larval form of *Echinococcus granulosus* (Dog tapeworm). Humans are the accidental intermediate host in the life cycle of hydatid disease. It is an endemic disease in the sheep and cattle raising countries of the Middle East, North Africa and South America. Although hydatid disease affects any organ or soft tissue, it is most frequently encountered in the liver (60–70%), lungs (10–40%), and rarely in the kidney, spleen, bone, thyroid, breast and pancreas. Clinical presentation varies according to the location of the cyst.

Splenic hydatid disease is a rare entity with less than 3% of the total incidence of Echinococcosis even in the endemic areas. Primary infestation of spleen takes place by the arterial route after the parasite has crossed the two filters i.e. hepatic and pulmonary. A retrograde venous route which bypasses the liver and lungs is also reported. Complications such as secondary infection, rupture into the peritoneal cavity or fistulisation to adjacent organs may be seen with splenic hydatid cyst. Traumatic or spontaneous rupture of hydatid cyst may cause systemic anaphylactic reaction which may be a life-threatening complication.

Case Report

A 65 year old female patient presented to us with pain in the left upper quadrant of the abdomen. There was left hypochondriac dull aching pain which did not radiate. There were no aggravating or relieving factors. Patient complained of nausea, occasional fever and weakness since one year. She had no history of jaundice, cough or respiratory complaints, abdominal trauma or weight loss and her past medical history was unremarkable. On examination, her vital parameters were within normal limits. Physical examination showed a distended abdomen with smooth surface in left hypochondriac and epigastric region. Mild tenderness was noted in the same region with no guarding or rigidity. There was no hepatomegaly or lymphadenopathy. Other systemic examination and routine laboratory investigations revealed no abnormalities. Chest radiograph was normal. Abdominal ultrasonography showed round, well defined, cystic lesion of approx. size 110 × 80 mm indenting left kidney and stomach.

Abdominal CT scan revealed enlarged spleen showing multi cystic lesion measuring 11 × 8 cm with multiple daughter cyst within. Spleen was indenting left kidney and displacing the stomach anteriorly and these findings were suggestive of hydatid cyst of spleen. There were no cysts in other abdominal viscera. Laparotomy was performed through a left subcostal incision. Surgical exploration revealed a hydatid cyst occupying whole the splenic parenchyma with only thin rim of splenic tissue present in inferior surface and at the notch. The mass was attached superiorly to left dome of diaphragm and postero inferiorly to the large bowel. A splenectomy was performed and the cyst was

resected en-bloc with the spleen. The rest of the abdominal organs including the liver were normal. To treat any potential contamination, the abdomen was locally washed with hypertonic saline solution (3% NaCl).

Histopathological examination showed spleen of size 18 × 13 × 6 cm on cut section, replaced completely by hydatid cyst with small amount of normal splenic tissue near the notch. Microscopic examination revealed laminated cyst wall with few daughter cysts underneath and necrosis, thus confirming the diagnosis of hydatid cyst of spleen. Post-operative course was uneventful and the patient was discharged on postoperative day 7. On follow-up, ultrasonography did not show any evidence of residual or recurrent cyst.

Discussion

Hydatid disease is a major problem worldwide, mostly in the sheep and cattle raising areas of the world. Primary hydatid disease of the spleen is a very rare condition. Usually splenic hydatid cysts are secondary, resulting from either spontaneous spread of cysts or occurring after surgeries involving hydatid cysts in other regions. Even in the endemic region, the frequency is reported to be 0.5 – 4% of abdominal hydatid diseases [8]. The most common sites of hydatid disease are the liver (60–70%), acting as a first filter followed by the lungs (10–40%), which acts as second filter. The other rare sites include spleen, kidney, thyroid, gall bladder, central nervous system and retroperitoneal region. Practically, any organ can be infested by the parasite. Splenic involvement is uncommon as the cyst embryos are trapped in the liver and lungs and only 15% of them enter systemic circulation. The eggs of the parasite escape the liver-lung barrier and cause primary infestation of spleen via the arterial route [4]. Splenic hydatidosis may also arise with retrograde spread of parasites through the portal and splenic veins bypassing the liver and lungs. Secondary splenic hydatid disease usually follows systemic dissemination or intra peritoneal spread following ruptured hepatic hydatid cyst.

The splenic hydatid cyst consists of three layers. The outermost adventitia (pericyst or pseudo cyst) is composed of compressed splenic tissue; a middle layer consisting of laminated membrane of friable ectocyst and an innermost germinal layer known as endocyst. The presentation of splenic hydatid disease can vary greatly and most of them are usually asymptomatic, solitary slow growing and incidentally diagnosed. The main symptoms associated with the disease are abdominal discomfort, pain and a palpable mass in the left upper quadrant. A study reported 38 cases of splenic hydatid cysts and abdominal pain was the most common symptom among these patients [2]. The complications of untreated splenic hydatid cyst are mainly secondary infection, acute abdomen and inflammation, adhesions with nearby organs such as kidney, diaphragm and stomach, compression of other viscera, intra-abdominal rupture and fistulization to the bowel,

mainly colon causing severe pain and may prove to be life-threatening. Teke et al. reported a splenic hydatid cyst perforating into the left colon and causing massive gastrointestinal bleeding^[3,4]. Other reports include rupture of splenic hydatid cyst into the thorax leading to spleno-thoracic fistula and severe anaphylactic reactions leading to fever, pruritus, dyspnoea, stridor and edema of the face. Cystic growth may cause compression of the segmentary vessels of spleen resulting in extensive pericystic splenic atrophy and the hydatid cyst may entirely replace the splenic parenchyma. The differential diagnosis of splenic hydatid cyst includes cystic lesions of spleen such as epidermoid cysts, hematomas, splenic abscess, post-traumatic pseudo cyst, neoplasms like lymphangioma and haemangioma. Preoperative diagnosis becomes difficult due to the similarity of presenting symptoms and radiological findings, to those of the other, more commonly encountered lesions of the spleen. The Casoni skin test is a sensitive test but not specific. On abdominal radiograph crumpled eggshell-like calcifications in the splenic area suggests splenic hydatidosis. Ultrasonography and CT scan are the major diagnostic tools for splenic hydatid cyst. Serological tests are also sensitive and specific for Echinococcosis. Owing to the risk of spontaneous or traumatic rupture and the limited efficacy of drug therapy, surgical approach is still accepted as standard treatment for managing hydatid disease. The standard treatment is splenectomy as complete resection removes all parasitic and pericystic tissues^[5]. Total or partial splenectomy, cyst enucleation and deroofting with omentoplasty are the various other preferred surgical techniques for treating splenic hydatid disease. The splenic cyst with adhesions or infiltration to nearby organ should be treated by total splenectomy as was done in our case. However, in cases of infected splenic cyst or metastatic implantations to adjacent organs, or if the location and size do not allow safe resection, conservative surgical techniques such as partial splenectomy, deroofting with omentoplasty, cyst enucleation or external drainage may be used^[6]. Attempts are usually made to conserve the spleen, so as to avoid opportunistic post splenectomy infections (OPSI)^[7]. But partial resection is usually avoided as it carries a risk of poor vascular control while deroofting the cyst wall leaves behind a residual cavity which carries the risk of postoperative infection. For the above reasons and also the possibility of multiple cysts, total splenectomy should be employed as the method of choice, especially in cases where there is presence of a communication between the spleen and nearby organs such as stomach, diaphragm or colon. Laparoscopic splenectomy has also been advocated for uncomplicated splenic hydatid cyst. Newer methods include puncture-aspiration-injection-reaspiration (PAIR) technique using hypertonic saline solution before opening the cavities which tends to kill the daughter cysts.

In the postoperative period, Medical treatment is the mainstay of treatment. Anti-helminthic drug therapy with Albendazole 10–15 mg/kg/day for one month or Mebendazole 40–50 mg/kg/day for 3–6 months, in addition to Praziquantel 40 mg/kg/week for 2 weeks pre and postoperatively to reduce the chances of anaphylactic shock and decrease the tension in the cyst wall are used. Albendazole is an effective adjuvant therapy in the treatment of hydatid disease lowering the chances of recurrence. Gil-Grande et al reported that albendazole sterilizes up to 72.3% of cysts by the end of the first month and 94% at the end of 3 months of treatment.

In our case we performed a total splenectomy after which the patient was given prophylactic vaccination against Streptococcus pneumoniae, Haemophilus influenza type b and Neisseria meningitidis, and was started on post-operative course of anti-helminthic drug. No post-splenectomy infection was encountered. Complete and aggressive surgical resection is the gold standard treatment for patients with hydatid cysts with the aim to remove all parasitic and pericystic tissues.

Conclusion

In conclusion, splenic hydatid disease is an infrequently encountered condition and should be considered in the differential diagnosis of all cystic masses in the abdomen/spleen, especially in the geographical regions where the disease is endemic. Its early diagnosis may become a challenging surgical problem and hence the pre-operative evaluation should be carried out carefully. CT scan is the most sensitive investigation for making a diagnosis. Although the management must be individualized for each patient, surgical resection (total splenectomy) is the best curative procedure following which medical treatment is necessary to ensure complete healing.

Fig1. Gross specimen showing spleen of size 18 × 13 × 6 cm on cut section, replaced completely by hydatid cyst with small amount of normal splenic tissue near the notch.



Fig2. CT Image showing multi cystic lesion in the spleen measuring 11 × 8 cm with multiple daughter cyst within.



References

- [1] Dontigny L, Mercier C, Pagé A, et al. An unusual case of hydatid cyst. *Can J Surg* 1976; 19: 23-5.
- [2] Tarcoveanu E, Plesa A, Danila N, Lupascu C, Cotea E, Negru R. Splenic hydatid cyst. Observations upon 38 cases of splenic echinococcosis. *Revista Medico-Chirurgicala a Societatii de Medici Si Naturalisti Din Iasi* 2002; 107: 311–5.
- [3] Lippitt WH, Akhavan T, Caplan GE. Epidermoid cyst of the spleen with rupture and inflammation. *Archives of Surgery* 1967. Jul; 95 (1):74–8.
- [4] Teke Z, Yagci AB, Atalay AO, Kabay B. Splenic hydatid cyst perforating into the colon manifesting as massive lower gastrointestinal bleeding: an unusual presentation of disseminated abdominal echinococcosis. *Singapore Medical Journal* 2008; 49: 113–6.
- [5] Hoffman E. Non-parasitic splenic cysts. *The American Journal of Surgery* 1957; 93(5):765–70.
- [6] Berrada S, Ridai M, Mokhtari M. Hydatid cysts of the spleen splenectomy or conservative surgery. *Ann Chir* 199; 45: 434–6.
- [7] Amatzidis K, Papaziogas B, Mirelis C, Pavlidis T, Papaziogas T. Splenectomy versus spleen-preserving surgery for splenic echinococcosis. *Digestive Surgery* 2003; 20: 527–31.
- [8] Celebi S, Basaranoglu M, Karaaslan H, et al. A splenic hydatid cyst case presented with lumbar pain. *Intern Med* 2006; 45: 1023–4.
- [9] Kiresi DA, Karabacakoglu A, Odev K, et al. Uncommon locations of hydatid cysts. *Acta Radiol* 2003; 44: 622-36.
- [10] Pedrosa I, Saiz A, Arrazola J, et al. Hydatid disease: radiologic and pathologic features and complications. *Radiographics* 2000; 20: 795-817.
- [11] Durgun V, Kapan S, Kapan M, et al. Primary splenic hydatidosis. *Dig Surg* 2003; 20: 38-41.