



Case Report

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Giant primary hydatid cyst of spleen: rare case report with brief review of literature

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Abstract

Primary extrahepatic hydatid cysts are rare, and primary splenic hydatid cysts even more so, constituting 2% to 3.5% of all hydatid cysts. We did a retrospective study of four years from Jan 2011 to Dec 2014 and only 24 cases of hydatid cysts were recorded. There was only one case of splenic hydatid cyst. Diagnosis was made with the help imaging modalities like ultrasonography and/or contrast-enhanced computed tomography. Enzyme linked immunosorbent assay for hydatid antibodies tested were done to confirm the diagnosis. Surgery was the main stay of treatment after proper sterilization of cyst cavity.

Keywords: Computed tomography, Hydatid disease, Splenic hydatid, Ultrasonography.

INTRODUCTION

Hydatid disease is an ancient disease and has a worldwide distribution. It is more common in sheep and cattle raising areas of the world.^[1] Humans contract the disease by ingesting highly infective eggs of adult echinococcus harbouring in the small intestine of the definitive hosts like dogs and canine animals. The liver acts as the first filter in trapping the embryos which then develop into hydatid cysts in 55 – 70% cases, followed by the lungs being 2nd filter in 18-35% cases. Incidence of unusual sites is about 8-10% and various sites are like spleen, psoas muscle, pelvic cavity, peritoneum, mesentery, brain, kidneys, bones, muscles and soft tissues. Hydatid cysts involving the spleen are rare (2.5%). Splenic hydatid, being a rare entity, can occur primarily or in association with hepatic, pulmonary or multi-organ hydatidosis.^[2] We report a case of splenic hydatid cyst due to rarity.

CASE REPORT

16 years old unmarried girl presented with 2 months history of pain and heaviness in the left hypochondrium. The pain was localized, gradual in onset, moderate in intensity, dull aching in character, associated with a feeling of heaviness and mass in upper abdomen. The rest of the systemic review was normal. Physical examination revealed a blood pressure of 110/ 70 mmHg, pulse 80 beats/min with mild anemia. On abdominal examination, there was a tender, irregular soft mass moving with respiration and occupying whole of the left hypochondrium about 6-7 cm from Lt. costal margin. A clinical diagnosis of cystic lesion of spleen was made. On investigations, TLC: was raised to 10300; ESR: 14; Eosinophil count was high (6 %). Indirect haemagglutination test for echinococcus was positive. X-Ray chest was normal. Ultrasonography examination abdomen showed a cyst in spleen (Fig-1) and liver with rest of abdominal organs were normal. Computed enhanced tomography (CECT) confirmed the diagnosis and it was 11.26 cm (AP) X 10.7 cm (ML) in size replacing almost whole of spleen (Fig-2). Exploratory laparotomy done through left sub costal incision revealed a huge splenic cyst occupying the lower pole. Scolidol agent 10% Betadine was installed in the cyst and this process was repeated four times. Then deroofting and pericystectomy done up to normal splenic tissue. Rest of the abdominal viscera including liver were normal. Abdomen was closed in layers without any drain. Tab. albendazole 10-15 mg/kg/day was continued, which she had already been taking preoperatively, for the last one month. The patient was discharged on 8th post-operative day in a satisfactory condition.

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Figure 1: Ultrasound showing splenic hydatid cyst



Figure 2: CECT showing huge hydatid cyst of spleen

DISCUSSION

Hydatid disease is endemic in Middle East as well as common in India, Africa, South America, New Zealand, Australia, Turkey and South Europe. Splenic cyst are classified as true cysts (primary) or pseudocysts (secondary) based on the presence of an epithelial lining. True cysts can be further subdivided into parasitic (caused by Echinococcus) and nonparasitic.^[3] Nonparasitic true cysts are congenital or neoplastic. Congenital cysts can be epidermoid, dermoid, or endodermoid, present at a young age (children and young adults) and are commonly located in the upper pole of the spleen. CA 19-9 and CEA levels are elevated in the epidermoid cyst's contents and in the patient's serum. Pseudocysts are believed to develop after post-traumatic intraparenchymal or subcapsular splenic haematoma and occasionally after splenic infarcts or infections. Secondary cysts account for 75% of all nonparasitic splenic cysts.^[3] The frequent use of abdominal imaging and the increasingly successful non-operative management of splenic injuries contribute to a rise in the incidence of nonparasitic splenic cysts, which otherwise are considered to be rare. Throughout the world, over two-thirds of the splenic cysts are parasitic hydatid cysts caused by Taenia Echinococcus. The differential diagnosis for splenic hydatid cysts includes other splenic cystic lesions such as epidermoid cysts, pseudocysts, splenic abscesses, hematomas and cystic neoplasms of the spleen.^[4]

Echinococcus granulosus is a member of the order Cestoda (flatworm) and family Taenia. Man is an accidental intermediate host, as entry of the larval forms into humans represents an end stage in its life cycle. Consumption of contaminated vegetables or meat, which are not washed or scrubbed free of eggs, exposes man to the larval forms. An alternate mode of entry is direct contact with dogs, whose fur has the eggs sticking onto it. Once in the intestine of man, these embryonic forms enter the portal circulation and can spread to a variety of organs, including liver, lungs, pancreas, spleen, etc.

About 10-15% of embryos escape from liver and lung and filters into general circulation. A small fraction of these escaped embryos settle down in the spleen. The cyst may settle in the interior of the spleen or at its periphery under the capsule. Infection is usually acquired in childhood but they mostly remain asymptomatic. The cyst grows at a rate of 0.3-1 cm per year and it may take 5-20 years to grow into a sufficient size (3 – 35 cms) to cause symptoms of a constant abdominal pain and a visible/palpable swelling in the abdomen.^[5] The incidence of hydatid cysts of spleen varies widely in sheep raising countries. In India the incidence of splenic hydatid cysts reported by different workers from different cities is as shown in Table 1.^[5]

Table 1: Incidence of hydatid cyst of spleen in India

Area	Incidence (%)
Central India	6.30
Delhi	4.20
Srinagar	4.10
Indore	3.30
Pondicherry	2.70
Ahmadabad	2.00

The disease affects all age groups and both sexes with equal frequency. Diagnosis is usually established incidentally during investigation of unrelated symptoms. When the cyst reaches an advanced size, the patient presents with a painless mass in the left hypochondrium. Some patients may present with complications such as infection of the cyst, rupture of the cyst into the peritoneal cavity, fistula formation into hollow viscera like colon or stomach.^[6] Moderate eosinophilia of 6% or more is usually present. There are several serological tests to diagnose hydatidosis; like serum immunoelectrophoresis which is currently the most reliable, with a sensitivity of approximately 90%, indirect haemagglutination has a sensitivity of 85%, ELISA and western blot analysis have also been used, in addition to eosinophilia, high IgE and IgM. The Casoni skin test is sensitive but not specific. Serologic tests are helpful for the diagnosis, screening and post-op follow-up for recurrence.

Preoperative diagnosis of this infection is mandatory as one of the common complications of hydatid disease is cyst rupture after trauma or spontaneous rupture as a result of increased intracystic pressure. Splenic hydatid cyst can rupture into a hollow viscous, into the pleural cavity through diaphragm, or directly into the peritoneal cavity. Cyst that rupture into the peritoneal cavity may cause peritoneal irritation, urticaria, anaphylaxis, and death. Therefore, hydatid cyst rupture needs immediate surgical intervention.^[7]

Since the condition closely resembles a soft tissue tumor on clinical examination, pre-operative radiological diagnosis is very important to avoid a biopsy. Plain films are usually incapable of showing cysts within a soft tissue. Preoperative diagnosis of hydatid cysts can be made on ultrasound and confirmed by Computed Tomography (CT) scan (94-96% and 100% sensitivity respectively). Ultrasonography is helpful in detecting calcification of the cyst wall, presence of daughter cysts, cystic membranes, septa or hydatid sand.^[8] In the present case ultrasound scan showed a large splenic cyst. C.T Scan added to the diagnostic accuracy by showing size, shape and extent of the lesion.

Treatment strategies include PAIR (puncture, aspiration, injection, and reaspiration) which was developed at the beginning of 1980s has proved to be successful in a variety of selected indications that have been reviewed by WHO recommendations.^[9] Today, percutaneous

treatment of liver hydatid cysts, a safe, easily applicable, and well-tolerated method, has become the most effective and reliable treatment procedure in most cases if the hydatid cyst is viable. In patients treated with the percutaneous treatment technique, a decrease in the dimensions of the cyst, solidification of the cyst contents, and irregularity in the walls of cysts are signs suggestive of cure.^[5]

Until the early 1970, open splenectomy was the standard treatment for benign splenic disorders, when fatal postsplenectomy sepsis was widely recognised. Post Splenectomy infection (OPSI) syndrome comprises fulminating bacteraemia, disseminated intravascular coagulation, multiple organs failure, severe hypoglycaemia and rapid death. Its incidence is about 0.9 to 60% and mortality rate is about 50%.^[5] So, tissue saving, partial splenectomy was introduced to reduce the chances of postsplenectomy sepsis, initially for trauma and later for most benign splenic diseases, including non-parasitic cysts, and parasitic hydatid cysts. Spleen preservation should always be done particularly in children. In the minimal invasive surgery era, laparoscopic is possible for benign splenic cyst and is also superior to open splenectomy. These days partial splenectomy for non-parasitic cysts is done laparoscopically. Possibility of anaphylactic reaction and recurrence due to spillage were the main concern with laparoscopy surgery. Despite all precautions, incidence of spillage of scolex-rich fluid during surgery is about 5%–10%, and this does not necessarily lead to dissemination. Recurrence after surgery was reported in up to 18% of cases, which may be due to incomplete removal, spillage, or growth of small occult cysts that were missed initially. Laparoscopy has become gold standard in trained hands and enucleation of cyst with omentopexy can be done laparoscopically.^[10]

Medical treatment with albendazole is used in preoperative period to reduce the size and postoperatively to reduce the incidence of recurrence. It is treatment of choice in patients who are unfit for surgery. In our case we decided preoperatively to use albendazole because possibility of hydatidosis in other parts of the body could not be completely eliminated, despite all radiological diagnostic modalities. There is always possibility of occult perioperative leak so medical therapy is mandatory. Mebendazole and albendazole are a benzimidazole derivative. These drugs interfere with mechanisms of glucose absorption through the wall of the parasite.

According to the WHO guidelines for the treatment of hydatid disease, chemotherapy is indicated for inoperable patients and those with multiple cysts scattered in many organs where surgery can be ineffective or hazardous.

CONCLUSION

Hydatid cyst of spleen is rare but a high degree of suspicion should be there whenever splenic cyst is encountered in clinical practice. Preoperative diagnosis sometimes become challenging as radiological techniques is usually unable to add to the diagnostic accuracy. Hydatid disease should be considered in the differential diagnosis of all cystic masses in the spleen/(abdomen), especially in the endemic areas. If diagnosed early, this disease can be treated completely. In such cases, diagnostic laparoscopy followed by exploration and preoperative decisions are worth practicing.

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