



## CASUISTIC PAPER

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# Splenic hydatidosis with abdominal pain – a rare presentation in a developing nation

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## ABSTRACT

**Introduction.** Primary extrahepatic hydatid cysts are rare, and primary splenic hydatid cysts even rarer. Splenic hydatidosis constitutes 2% to 3.5% of all hydatid cysts.

**Aim.** To present a case report of splenic hydatidosis with abdominal pain.

**Description of the case.** We report here a case of isolated splenic hydatid cysts in a 23 year old female, who presented with dull dragging pain in the left hypochondrium. Diagnosis was made on computed tomography imaging of the abdomen and microscopic examination of the laminated hydatid cyst wall and supplemented with positive enzyme linked immunosorbent assay for hydatid antibodies.

**Conclusion.** The incidence of splenic involvement by hydatid cysts is very low. Man is an accidental intermediate host, as entry of the larval forms into humans represents an end stage in its life cycle. Until recently the gold standard treatment for splenic hydatidosis was splenectomy, as medical therapy seems to be ineffective. However, the last two decades have shown a tendency towards splenic conservative surgery in suitable cases, to reduce opportunistic post splenectomy infection.

**Keywords.** abdominal pain, histopathology, hydatidosis, spleen

## Introduction

The first description of splenic hydatid cyst was given by Berlot in 1790.<sup>1</sup> Hydatidosis also known as echinococcosis, is a parasitic infection of liver and other organs by flatworm, echinococcus. *Echinococcus granulosus* is the causative organism of hydatid cysts. Most hydatid cysts are acquired in childhood with a latent period of five to twenty years before the diagnosis.<sup>2</sup> The growth of hydatid cyst is very slow, as a very crude estimate of an increase in the diameter by about

two to three centimeters each year.<sup>3</sup> The rate of growth of hydatid cysts depends not only on immunologic relationship between the parasites and humans but also on the resistance offered by the enveloping structure. The incidence of hydatid cysts has decreased in the endemic areas due to enforcement of public health measures and livestock handling procedures. Various measures like, public education about the disease and its transmission, instructions for vigorous hand washing after contact with canine species, elimination of vegetables grown at

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ground level from the diet, and discontinuation of the practice of feeding the entrails of slaughtered animal to dogs have led to the decrease in the occurrence of hydatidosis.<sup>4</sup> A mean incidence of splenic hydatid cyst in India is 4.3%, with the disease affecting all age groups and both sexes with equal frequency.<sup>4</sup>

### Aim

To present a case report of splenic hydatidosis with abdominal pain.

### Description of the case

A 23-year-old woman presented to the general surgery clinic with dull aching pain in the left upper quadrant of the abdomen for the past 3 months. Physical examination revealed tenderness and an ill-defined firm mass on the left side. Hemogram showed mild eosinophilia. Abdominopelvic computed tomography (CT) scan showed a 10×9×5 cm loculated cyst presenting as an unenhanced hypodense mass with well-defined borders in the spleen (Figure 1).



**Fig. 1.** Abdominopelvic computed tomography (CT) scan showed a single 10 × 9 × 5 cm loculated cyst presenting as an unenhanced hypodense mass with well-defined borders in the spleen

A CT scan of his chest did not show any cysts. On exploration, a huge hydatid cyst in the spleen of size 10.5×8.7×6.1 cm, filling the left side of the abdomen was seen. The contents of the splenic hydatid cyst was aspirated and all the endocysts were removed, followed by splenectomy. On microscopic examination, the hydatid cyst wall showed the characteristic laminated adventitial layer adjacent the red and white pulp of the spleen (Figures 2, 3, 4 and 5). The postoperative period was uneventful, and she was discharged with albendazole treat-

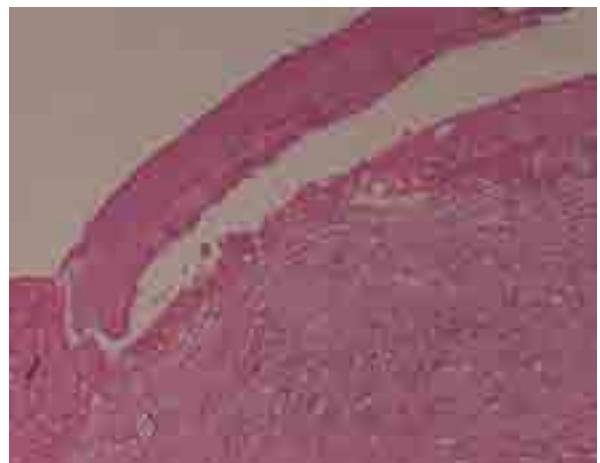
ment with a dose of 10 mg/kg/day for 6 months, on the postoperative day.



**Fig. 2.** Microscopic examination shows different sized lymphoid follicles in the splenic pulp with foci of the hydatid cyst wall with the characteristic laminated adventitial layer (10X) (H&E stain)



**Fig. 3.** Tissue section shows the hydatid cyst wall showed the characteristic laminated adventitial layer adjacent the red and white pulp of the spleen (10X) (H&E stain)



**Fig. 4.** Tissue section shows the hydatid cyst wall with the characteristic laminated adventitial layer (10X) (H&E stain)

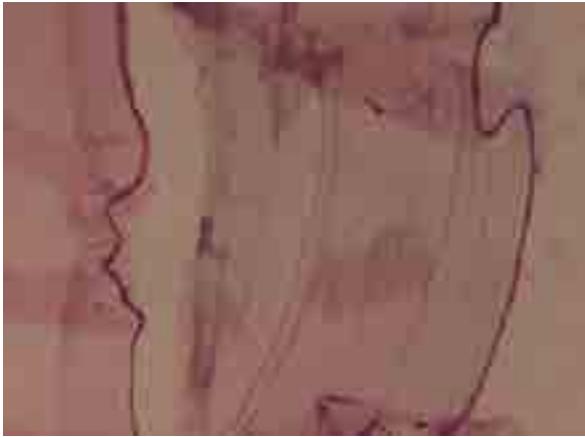


Fig. 5. High power of Figure 4 (40x) (H&E stain)

### Discussion

Hydatid cyst is the only parasitic cyst of the spleen.<sup>5</sup> In India, a mean incidence of splenic hydatidosis is 4.3%.<sup>5</sup> Liver is the most common site where hydatid cysts develop (70%), which acts as a first filter, followed by the lungs (15%), which acts as the second filter. No organ is immune to infestation by hydatid disease.<sup>6</sup> The rare sites of hydatid cysts include thyroid, omentum, pancreas, gall bladder, central nervous system and the kidney.<sup>7-9</sup>

Echinococcal cysts of the spleen usually grow very slowly and patients may be asymptomatic for 5-20 years before diagnosis. The symptomatology is usually mild with usual complaints of mild discomfort or pain in the left hypochondrium. The symptom complex produced by splenic hydatid cyst are mainly due to mechanical displacement and pressure effect on the adjacent organs.<sup>10</sup> Approximately 30% of splenic cysts are asymptomatic and detected incidentally. In addition 40% have no detectable physical signs.<sup>10,11</sup> Physical examination may reveal either a large palpable spleen or a hard, round and smooth mass, which follows the respiratory movements of the diaphragm.

Traub reported a splenic cyst causing hypertension by renal artery compression.<sup>10</sup> The splenic cyst can rupture and disseminate all over the peritoneal cavity. Other reported complications of splenic hydatid cysts are acute abdomen produced by splenic hydatid cysts coexistent with bilateral ovarian tumor, rupture of long standing splenic hydatid cyst into bronchial tree, traumatic rupture of splenic echinococcal cyst with anaphylactic shock.<sup>11,12</sup> Laboratory evaluation of patients with hydatid disease often yields non-specific data. Eosinophilia is not significant in endemic areas.<sup>4</sup>

A large battery of serological tests are available but their importance have been diminished by increased reliance upon modern imaging modalities like ultrasound (USG), computed tomography (CT) and Magnetic resonance imaging (MRI). Several serological tests are specific to hydatidosis and are used to con-

firm the diagnosis. Serum immunoelectrophoresis is currently the most reliable, with a sensitivity of approximately 90%.<sup>4</sup>

Complement fixation, enzyme linked immuno-sorbent assay (ELISA) and western blot analysis have also been used. The Casoni skin test is sensitive but not specific, and also remains positive for years after eradication of the organism. Plain radiology of abdomen can reveal a soft tissue shadow with or without calcification, displacement of left diaphragm upwards, stomach to the right and transverse colon with splenic flexure downwards. Celik et al have suggested displacement of neighbouring organs is characteristic of cyst of spleen rather than splenomegaly from other causes.<sup>13</sup> Ultrasound and Computed tomography alone or in combination established the definite diagnosis of splenic hydatid cysts in almost all the cases. These procedures can identify daughter cysts and hydatid sand, both of which are specific to echinococcal infestation.<sup>4</sup> Ultrasound is cost effective and particularly valuable for follow up screening. CT is more accurate than ultrasound in localizing and delineating extent of the cyst.<sup>14</sup> Postoperative histopathology will confirm the parasitic nature of this splenic cyst.

The development of echinococcal cysts in the spleen is uncommon because hexacanth embryos are usually trapped in the liver (first Lemman's filter) and/or lung (second Lemman's filter) but will be trapped in the splenic capillaries once in the systemic circulation.<sup>8</sup> Splenic echinococcosis may also arise by retrograde spread from the liver to the spleen via the hepatic portal and splenic veins in portal hypertension. The spleen may also be affected by rupture of a hepatic echinococcal cyst into the peritoneal cavity.<sup>1,5</sup>

Splenic echinococcosis needs to be differentiated from non-parasitic cysts, epidermoid cysts, haemangiomas, sarcomas, pseudocysts and tumours of the diaphragm, stomach, colon, left kidney or pancreas. The diagnosis is made by the history, physical examination, the presence of peripheral calcification or daughter cysts within a large cystic lesion or coexistent cystic lesions in the liver or other organs.<sup>13,15</sup>

The surgical procedures employed are total splenectomy and cyst enucleation and tube drainage of the cavity. The diagnosis of echinococcal cyst is confirmed by histopathology in all of the resected specimens. Medical treatment comprises of Mebendazole (60 mg/kg/day for 6-24 months) or Albendazole (10 mg/kg/day for 6 months). The treatment of hydatid cysts is principally surgical. However, pre- and post-operative 1-month courses of Albendazole and 2 weeks of Praziquantel should be considered in order to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall (thus reducing the risk of spillage during surgery) and to reduce the recurrence rate post-opera-

tively.<sup>4</sup> Intra-operatively, the use of hypertonic saline or 0.5% silver nitrate solutions before opening the cavities tends to kill the daughter cysts and therefore prevent further spread or anaphylactic reaction.<sup>4</sup>

## Conclusion

The incidence of splenic involvement by hydatid cysts is very low. Man is an accidental intermediate host, as entry of the larval forms into humans represents an end stage in its life cycle. Until recently the gold standard treatment for splenic hydatidosis was splenectomy, as medical therapy seems to be ineffective. However, the last two decades have shown a tendency towards splenic conservative surgery in suitable cases, to reduce opportunistic post splenectomy infection.

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