

# Recurrent Primary Isolated Hydatid Cyst of Spleen with Giant Splenic Lipoma Mimicking Liposarcoma: A Case Report

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## 1. Abstract

### 1.1. Background

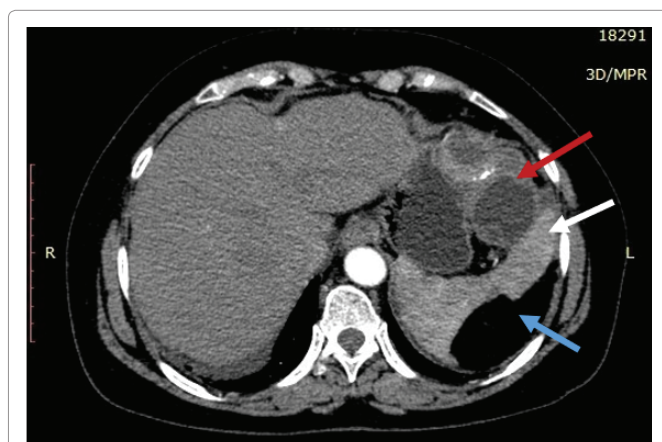
The incidence of splenic hydatid cyst is 1% and it comprises 0.5% - 4% of abdominal hydatid cyst. The recurrence rate of hydatid cyst is reported to be in the range of 4.6% - 22% in different literatures. Primary hydatid cyst of spleen is rare even in endemic areas, comprising only 2% of all splenic hydatid cyst. Concurrent recurrent isolated splenic hydatid cyst with giant splenic lipoma has never been reported in the literature till date.

### 1.2. Case Summary

50 year Female who had undergone enucleation of hydatid cyst of spleen 4.5 years back, presented with chief complaints of pain in left upper quadrant of abdomen for 2 weeks. Per abdominal examination revealed mild tenderness over left hypochondrium with no organomegaly or palpable lump. CECT abdomen and pelvis revealed hydatid cyst of spleen with lipoma or liposarcoma, for which total splenectomy was done.

### 1.3. Discussion

Microscopic spillage of viable parasite or failure to remove the cyst completely during index surgery are responsible for recurrence of hydatid cyst. Negative serology does not rule out hydatid cyst. Only 54.8% patients with early or inactive cyst stages show positive serological reaction. Similarly, the distinction between lipoma and well differentiated liposarcoma, radiologically, is a diagnostic dilemma.



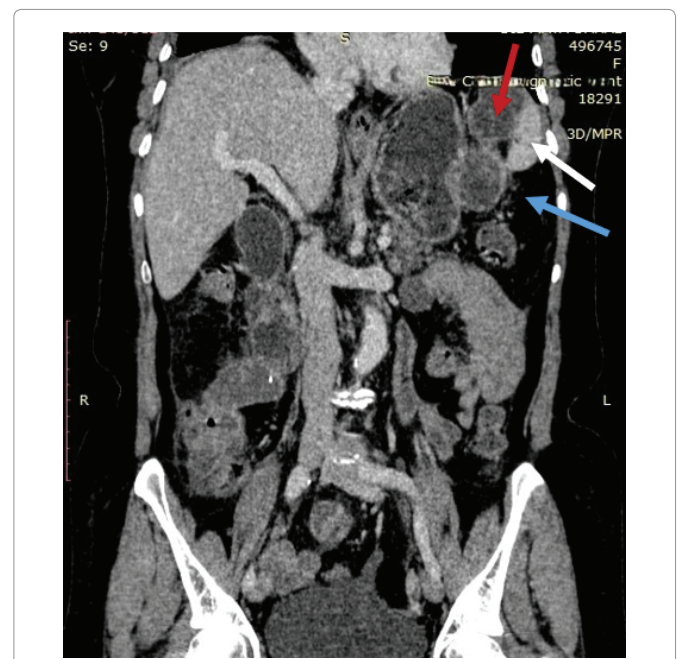
**Figure 1:** Axial view white arrow showing splenic tissue, red arrow showing well defined cystic lesion with few foci of calcification and hyperdense membrane/septa suggestive of hydatid cyst, blue arrow showing ill defined fat attenuating lesion on posterolateral surface of spleen suggestive of lipoma or well differentiated liposarcoma.

## 1.4. Conclusion

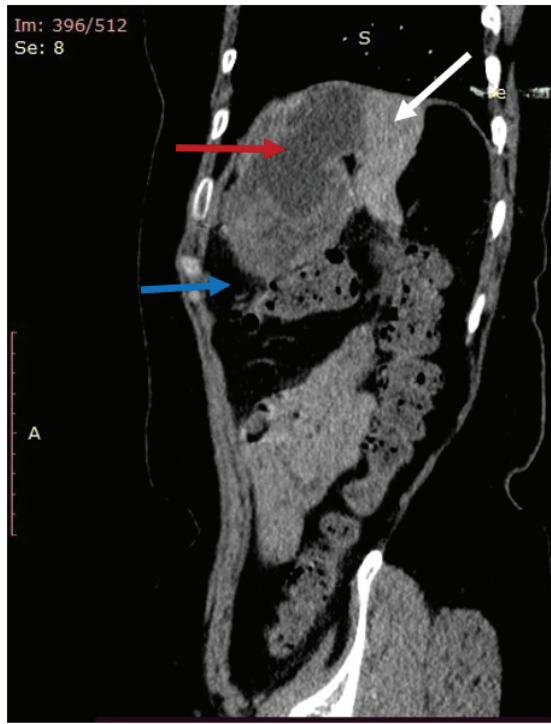
The treatment of splenic hydatid cyst can be either total splenectomy or spleen preserving procedures. It is recommended to continue albendazole for at least 1 month or mebendazole for at least 3 months after PAIR/surgery. Chemotherapy can be continued throughout life in inoperable cases or when only palliative resection is achieved.

## 2. Introduction

Hydatid cyst or Echinococcosis is a parasitic infection caused by larval stage of Echinococcus. Altogether, 3 types of echinococcosis, seen in human, are: cystic echinococcosis (CE) caused by *E. granulosus*, Alveolar Echinococcosis (AE) caused by *E. multilocularis* and Polycystic echinococcosis (PE) caused by *E. vogeli* and *E. oligarthus*. Out of these 3 forms, CE and AE are seen worldwide whereas PE is restricted to Central and South America [1,2]. Hydatid cyst can involve any organ in the body. The most common organ to be affected is liver (50-70%) followed by lungs (25%). Spleen is the third most common organ to be affected. The incidence of splenic hydatid cyst is 1% and it comprises 0.5% - 4% of



**Figure 2a:** Coronal view white arrow showing splenic tissue, red arrow showing well defined cystic lesion with few foci of calcification and hyperdense membrane/septa suggestive of hydatid cyst, blue arrow showing ill defined fat attenuating lesion on posterolateral surface of spleen suggestive of lipoma or well differentiated liposarcoma.



**Figure 2b:** Sagittal view white arrow showing splenic tissue, red arrow showing well defined cystic lesion with few foci of calcification and hyperdense membrane /septa suggestive of hydatid cyst , blue arrow showing ill defined fat attenuating lesion on postero lateral surface of spleen suggestive of lipoma or well differentiated liposarcoma.

abdominal hydatid cyst [3]. The first case of splenic hydatid was noticed during autopsy by Berlot in 1970 [4]. Splenic hydatid cyst can be either primary/isolated (cyst involving spleen only) or secondary (presence of hydatid cyst in spleen along with involvement of other organ) [5]. Primary hydatid cyst of spleen (as seen in our case) is rare even in endemic areas, comprising only 2% of all splenic hydatid cyst [6]. Rest 98% splenic hydatid cysts are secondary, due to systemic dissemination or intraperitoneal spread, most commonly from ruptured hepatic hydatid cyst [7]. The work has been reported in line with the SCARE criteria [8].

**3. Case Report**

50 year Female with known case of Hypertension and DM II under medication, presented with chief complaints of pain in left upper quadrant of abdomen for 2 weeks which was acute in onset, dull in character, non-radiating and mild to moderate in intensity. There were no history of nausea, vomiting, fever, yellowish discoloration of body, loss of appetite or significant weight loss. She underwent enucleation of splenic hydatid cyst 4.5 years back. Per abdominal examination revealed mild tenderness over left hypochondrium with no palpable mass or organomegaly. Routine baseline blood investigations were normal. On ultrasonography, 66cm x 64cm x 63cm cystic lesion was noted in left hypochondrium. On CECT abdomen and pelvis, well defined cystic lesion measuring 9cm x 8cm x 4.3cm with hyperdense non enhancing septa with few foci of calcification were noted, which was suggestive of hydatid cyst of spleen. The lesion was abutting medial surface of spleen laterally, greater curvature of stomach medially, left colon inferiorly and left diaphragm superiorly. Similarly, on posterolateral surface of spleen, 12cm x 9.6cm x 4cm ill defined fat attenuating lesion (-107HU) with thin septa was noted suggestive of lipoma or liposarcoma of spleen. ELISA for Echinococcal antibody was negative. Thus, total splenectomy was done (intra operative images could not be gathered due to technical issues). Along with initiation of albendazole, Hib, meningococcal and pneumococcal vaccination were given 2 weeks before splenectomy. After splenectomy, she was discharged on 5th postoperative day and was advised to continue albendazole for 1 more month.

**3.1. Pathological Findings**

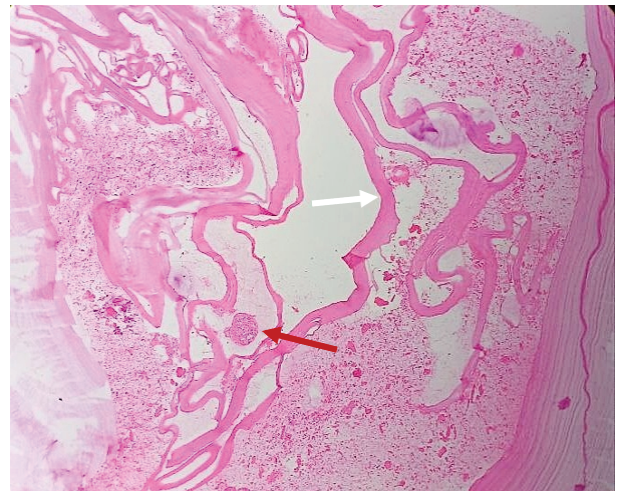
Grossly, the specimen consisted of two fragments. One portion,

representing part of the spleen, measured 7cm x 5cm x 3.5cm, while the other, a cystic structure filled with yellowish fluid, measured 12cm x 11cm x 3cm.

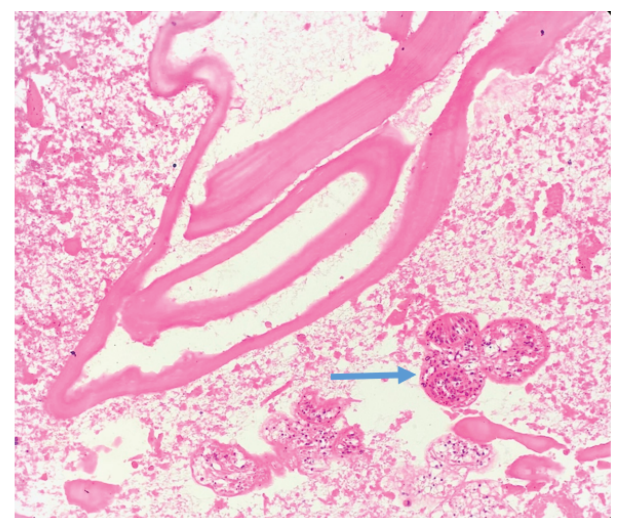
Histological examination revealed cystic structures lined by an innermost germinal layer, accompanied by an outer thick, avascular, eosinophilic, lamellated layer. The presence of scolexes and daughter cysts were also noted. Additionally, dense inflammatory infiltrates composed of neutrophils, lymphocytes, and plasma cells were observed. Thickened and congested blood vessels were also evident (as shown in Figure 3,4). Histological examination of the part of the spleen revealed normal splenic morphology. An additional specimen from the same patient comprised fibrofatty tissue measuring 11.5cm x 7.5cm x 3cm. The cut surface revealed homogenous yellowish area, and microscopic analysis demonstrated features characteristic of a lipoma (Figure 5). Histologically, the final diagnosis was splenic hydatid cyst with lipoma.

**4. Discussion**

About 30% of the patients are asymptomatic due to slower growth of cyst (0.3 - 2cm/year) [4,5]. Others may present with abdominal pain, abdominal lump, dyspnea (due to elevation of left diaphragm by cyst), constipation (due to compression of colon by cyst) or systemic hypertension (due to compression of renal artery by cyst) [9]. If the

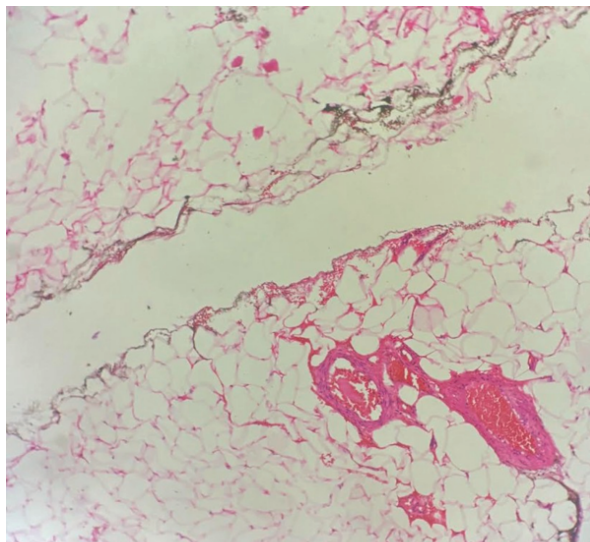


**Figure 3:** Splenic Hydatid cyst lined by an outer layer of avascular eosinophilic laminated chitinous layer (white arrow) with an innermost germinal layer and protoscolices (red arrow). (Hematoxylin and Eosin Stain, 4x).



**Figure 4:** Microscopic picture showing multiple daughter cysts and scolex containing multiple hooklets (blue arrow). (Hematoxylin and Eosin Stain, 40x).





**Figure 5:** Section from attached fibrofatty tissue showing mature adipocytes in lobules, individual cells having congested blood vessels and cytoplasmic vacuolation with nuclei pushed towards periphery suggestive of lipoma.

cyst compresses hilar vessels of spleen, there will be splenic atrophy eventually leading to complete replacement of splenic parenchyma. In case of chronic pericystic inflammation, there will be adhesion and hence fistulization of cyst with surrounding organs like stomach, left colon, pancreas, left bronchus or left kidney [10].

Negative serology (as seen in our case) does not rule out hydatid cyst. Serological test like ELISA can be negative due to ability of Echinococcus antigen to inhibit B cell activity and its proliferation. Only 54.8 % patients with early or inactive cyst stages show positive serological reaction. The sensitivity of ELISA increases with active cyst stages CE2 and CE3 [11]. Antibody detection by ELISA or immunoblotting is more sensitive than detecting Echinococcal antigen [12]. Even isolated lipoma of spleen is rare [13]. Here we encountered a case of recurrent isolated primary hydatid cyst of spleen along with giant lipoma of spleen, which has been never reported in the literature. Although the distinction between lipoma and well differentiated liposarcoma is a diagnostic dilemma [14], the radiological characteristics like heterogeneous fatty attenuation, presence of thick septa with marked enhancement, large size, nodular or globular areas of non-adipose tissue occupying more than 25 % of lesion favour the diagnosis of liposarcoma [14]. However presence of thin septa doesn't exclude liposarcoma. In our case, radiological characteristics like thin septa and absence of solid or enhancing component within the fat attenuating lesion were pointing towards the diagnosis of lipoma whereas large size (>10 cm in size) of fat attenuating lesion and advanced age of patient (50 years of age) were misleading the diagnosis as malignant liposarcoma. Recurrence of hydatid cyst (as seen in our case) is one of the major concern in the management of hydatid cyst. The recurrence rate is reported to be in the range of 4.6 % - 22 % in different literatures. Microscopic spillage of viable parasite or failure to remove the cyst completely during index surgery are responsible for recurrence [16]. Treatment options available for splenic hydatid cysts are total splenectomy and conservative spleen preserving procedures (partial resection of cyst wall/deroofing followed by omentoplasty, cyst enucleation, partial splenectomy, internal drainage with cystojejunal anastomosis, percutaneous drainage by PAIR technique) [17,18]. Partial resection of cyst or cyst enucleation carries higher risk of intraoperative rupture due to thin and fragile membrane of hydatid cyst (as compared to hepatic hydatid cyst which has thicker membrane) [4]. Also, there is high probability of leaving residual cyst wall and hence recurrence (as seen in our case) whereas partial splenectomy causes increased risk of post-operative hemorrhage. The only drawback of total splenectomy is post splenectomy sepsis which can be avoided by Hib and meningococcus vaccination done 2 weeks before or after splenectomy [17]. Conservative procedures are preferred in children (as they are at high

risk of developing post splenectomy sepsis and mortality rate exceeding 50 % [4]) and in adult when the cysts are small, inactive, superficial, lying at the upper or lower poles of spleen or associated with severe adhesion. PAIR (puncture, aspiration, injection and reaspiration) technique is safe for type I or II splenic hydatid cyst with diameter < 5 cm, patients who are not fit for anesthesia or refuse surgery [18]. In a study conducted by Atmatzidis et al., during a period of 25 years, 19 patients of splenic hydatid cyst were operated and it was concluded that there were no significant difference in the recurrence rate following splenectomy (12%) vs spleen preserving surgery (14%). Thus spleen preservation should be done if possible and the procedure of choice should be selected on the basis of size and location of cyst [10]. According to WHO guidelines, in case of Cystic Echinococcus, it is recommended to initiate chemotherapy 4 days before PAIR and 1 month before surgery. Similarly, it is recommended to continue albendazole for at least 1 month or mebendazole for at least 3 months after PAIR/surgery. Chemotherapy can be continued throughout life in inoperable cases or when only palliative resection is achieved [4]. The recommended dose of albendazole is 10-15mg/kg/day in 2 divided dose (practically 400mg BD in adult) [2].

### 5. Conclusion

Hydatid cyst of spleen is rare with incidence of 1%. Primary hydatid cyst of spleen is extremely rare comprising only 2% of all splenic hydatid cyst. It is often difficult to differentiate between lipoma and liposarcoma radiologically. The treatment of splenic hydatid cyst can be either total splenectomy or spleen preserving procedures.

### 6. Highlights for Review

1. Primary hydatid cyst of spleen is rare, comprising only 2% of all splenic hydatid cyst.
2. Even isolated lipoma of spleen is rare.
3. Recurrent isolated hydatid cyst of spleen along with giant lipoma of spleen has never been reported in the literature.
4. It is often difficult to differentiate between lipoma and liposarcoma radiologically.
5. The recurrence rate of hydatid cyst is reported to be in the range of 4.6% - 22% in different literatures.

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