DOI: 10.7860/IJARS/2023/56730.2854

Case Report

Surgery Section

Splenic Hydatid Cyst- A Case Report

ASHISH B JATALE[1], SANJANA IBRAHIMPUR[2], SAGAR R AMBRE[3], JAYASHRI S PANDYA[4]

ABSTRACT

Asymptomatic hydatid cyst presents as an incidental finding. It is caused by *Echinococcus granulosus*. The liver is known to be the involved organ, while the spleen is rarely involved. Hydatid cyst of the spleen should be considered as one of the differential diagnoses in patients presenting with left hypochondriac pain.This poses a clinical challenge. The decision on conservation or surgery is also a dilemma. To decrease incidence of Overwhelming Postsplenectomy Infection (OPSI) for elective splenectomy, vaccination protocol should be followed. It is important for the clinician to bear in mind the possibility of incidental asymptomatic splenic hydatid cyst and management protocols. The authors, hereby report, a case of a 26-year-old lady with complaint of left hypochondriac pain, diagnosed with Ultrasonography (USG) and Contrast Enhanced Computed Tomography (CECT) scan as splenic hydatid cyst. The patient underwent splenectomy after completion of vaccination to avoid OPSI. At six month follow-up, the patient was asymptomatic.

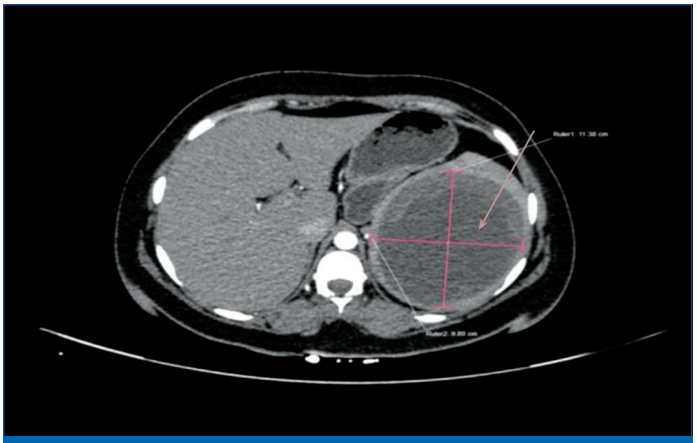
Keywords: Abdominal pain, Left hypochondriac lump, Overwhelming postsplenectomy infection

CASE REPORT

A 26-year-old female with complaint of left hypochondriac pain for 24 months duration was admitted to a tertiary care centre in Mumbai. The patient had pain at intervals, intermittent, non progressing, non radiating, with no aggravating and relieving factors. The patient also had abdominal fullness and early satiety. There was no history of fever, swelling, previous operation and any pet at home. Bowel and bladder habits were normal. General and clinical examination was normal. On abdominal examination, an ill-defined palpable lump was present in the left hypochondriac region of approximate size 6×6 cm, moving with respiration, which appeared cystic in consistency. As the patient had presented with pain in left hypochondrium, fullness and early satiety, clinical diagnosis of splenic enlargement was done and Ultrasonography (USG) was done to confirm the same. Ultrasound of the abdomen revealed enlarged spleen with well-defined rounded anechoic lesion seen in the upper pole of spleen, measuring 9×9×8 cm suggestive of splenic abscess, hydatid cyst, epidermoid cyst, haematoma, pseudocyst, neoplasm. The cyst was anechoic in nature so, could be either a parasitic cyst (most common hydatid) or non parasitic like an epidermoid cyst or a pseudocyst (majority have a history of trauma). Blood investigations were within normal limits.

Contrast Enhanced Computed Tomography (CECT) scan of abdomen and pelvis was done for further evaluation of lump which revealed well-defined, thick-walled cystic lesion measuring 11×9×11 cm with non enhancing thick membrane in non dependent position within a small eccentric peripherally placed daughter cyst. The cyst was seen to cause mass effect by pushing left kidney inferolaterally, and displacing splenic vein anteriorly, diaphragm superiorly, distal body and tail of pancreas and splenic artery inferiorly [Table/Fig-1]. This was suggestive of hydatid cyst type-2. High resolution CT (HRCT) of the thorax was done to rule out lung hydatid.

In view of elective splenectomy, the patient was prophylactically vaccinated against capsulated organism (*Pneumococcal, Meningococcal* and *Haemophilus influenzae*) to prevent Overwhelming Postsplenectomy Infection (OPSI). The patient was prescribed tablet albendazole for four days prior to surgery to reduce tension, stabilise cyst and decrease chances of rupture during surgery. Elective exploratory laparotomy was done for splenectomy and excised tissue was sent for histopathology.

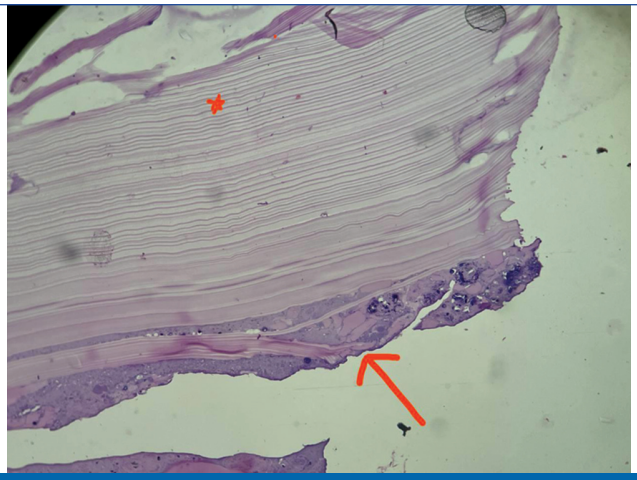


[Table/Fig-1]: CECT of the abdomen showing a large splenic hydatid cyst (arrow).

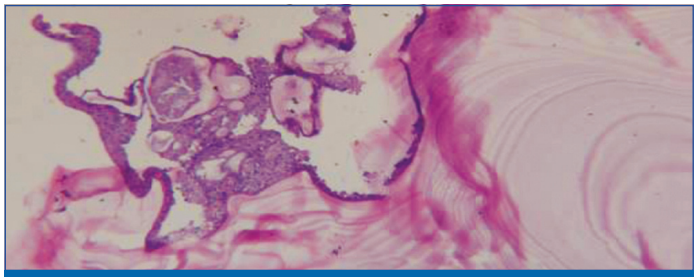
On histopathological examination, grossly, the lesion was single 17×17×7.5 cm multiloculated cystic mass occupying the entire spleen, with a thin small rim of splenic tissue present at the periphery. On cut section, clear liquid (hydatid fluid) approximately 100 mL was present in the cyst. Ectocyst had a tender coconut appearance. Endocyst showed scolex and daughter cyst [Table/Fig-2]. Histopathology showed lamellated ectocyst (star) and granular endocyst (arrow) [Table/Fig-3] and endocyst cellular lining with granular contents [Table/Fig-4].

The postoperative period was uneventful and the patient recovered well. In the postoperative period, the patient continued on tablet albendazole for a month. The patient was followed-up after six months with normal sonography.

[Table/Fig-2]: A gross cut section of a specimen shows a splenic hydatid cyst.



[Table/Fig-3]: Microphotograph of hydatid cyst (10x H&E stain).



[Table/Fig-4]: Microphotograph of hydatid cyst (40x H&E stain).

DISCUSSION

Hydatid disease is a zoonotic disease caused by *E. granulosus* [1]. Hydatid cyst commonly affects the liver with incidence of 50% to 75%, followed by lung with a 5% to 25% incidence [2-4]. Isolated splenic involvement is rare with worldwide incidence of 0.5-4% [4]. In India, in central part, the incidence is highest amounting to 2.5% [5].

The mechanism of primary splenic involvement is either through the splenic artery after bypassing the lung and liver or through the splenic vein by retrograde involvement [4]. Hydatid disease is more common in sheep-grazing area. Man and sheep are intermediate host where as dog is definitive host. After ingestion of egg, larva released from egg penetrate bowel mucosa to get access to portal circulation and spread to other organs [6]. The patient may be asymptomatic for long time or present with non specific dull aching left hypochondriac pain or mass, hypersplenism, dyspepsia heart burn, constipation and left sided portal hypertension rupture or fistula formation to colon [4-7].

Preoperatively, serological test like Enzyme Linked Immunosorbent Assay (ELISA), Immunoelectrophorosis (90-95% sensitivity), and indirect haemagglutination test (85% sensitivity) when used with radiological imaging techniques can give diagnosis of splenic hydatid in 90% of the cases [4]. Arc 5 capron on electrophoresis is the most reliable test for diagnosis of hydatidosis [8].

Differentials for left hypochondriac pain and mass are splenic abscess, hydatid cyst, epidermoid cyst, haematoma, pseudocyst and neoplasm. The cyst was anechoic in nature so, could be either a parasitic cyst (most common hydatid) or non parasitic like an epidermoid cyst or a pseudocyst (majority have history of trauma).

USG is a primary investigation with a sensitivity of approximately 90-95% that is not specific but useful in detecting daughter cysts. USG findings are that of benign cyst. It may be mistaken for an abscess or neoplasm due to mixed echogenicity. Some classical signs seen on USG are snake/serpentine sign, spin/whirl sign, double line sign, and water lily sign [4]. Due to mixed echogenicity of the membrane, scolices and hydatid sand, sonography shows higher echogenic pattern due to large acoustic impedance difference between the intracystic contents [8]. If egg shell calcification is seen on plain radiography in the left hypochondriac region, it can indicate splenic hydatidosis [5].

CT has higher sensitivity than USG and is useful for the determination of the size, number, and location of cysts [5]. CT attenuation depends upon intracystic contents. High Resolution Computed Tomography (HRCT) attenuation values are seen when there is presence of intracystic debris, hydatid sand, inflammatory cells and non scan [8].

Magnetic Resonance Imaging (MRI) is an important imaging technique for diagnosis and evaluation of hydatid disease. Patients with proven or suspected hydatid cyst MRI is not mandatory and not used as the first investigation in all cases of splenic hydatid cyst. Recently, for diagnosis and evaluation of focal splenic disease, CECT and USG are investigations of choice [4,8,9].

A combination of medical and surgical line of management is applied to treat human hydatidosis. In medical line of treatment, drugs used have parasitostatic action and not parasiticidal action, which results in low cure rate [5]. In surgery, open and laproscopic approach are employed. When there is small sized and superficially located cyst without any complication, laproscopic surgery is predefined. Though success rate of laparoscopy and open surgery are similar, if cysts are multiple, large in size, located deep in the organ, infected or ruptured, open surgery is the choice [10].

Due to the risk of spontaneous or traumatic rupture of the cyst, leading to anaphylaxis, splenectomy is preferred in cases with large hydatid cyst as splenic parenchyma is reduced significantly due to pressure atrophy. Splenectomy is associated with gastrointestinal complications like fistula formation to adjacent organs, rupture into the peritoneal cavity, and upper or lower gastrointestinal bleeding, gastric injury. Other complications include haemorrhage, pancreatitis, secondary infection, anaphylaxis and OPSI [9].

To decrease the chances of OPSI, patients are vaccinated for the same before undergoing surgery when posted electively. Though surgery cures the disease and is the treatment of choice in isolated splenic hydatid cyst, secondary *Echinococcus* occurs in 2-21% of cases due to spillage. Recurrence is not seen after complete resection of intact cyst [10].

The World Health Organisation (WHO) recommends one month prior or four days preoperative treatment with tablet albendazole to reduce intraoperative and postoperative complications [10]. The patient is to be monitored for at least 3 years postsurgery and patients who are managed with medical line of treatment with USG every 6 months [10-12]. [Table/Fig-5] shows the findings of a few similar published cases from the literature [3,6,13-19].

Author	Year	Number of cases	Clinical findings/symptoms	Radiological	Plan
Kumar P et al.	2016	1	Suggestive of peptic ulcer disease in left hypochondriac region	Suggestive of hydatid cysts	Laparoscopic splenectomy
Vezakis A et al.	2012	2	Abdominal pain with palpable abdominal mass	Ultrasound suggested enlarged spleen. Abdominal CT showed a splenic calcified hydatid cyst	1 case laparoscopic splenectomy 1 case open splenectomy
Malik AA and ul Bari S	2019	8	-	-	Splenectomy
Hepgul G et al.	2010	1	-	-	Splenectomy
Karakaya K	2007	2	-	-	Splenectomy

Durgun V et al.	2003	14	-	-	Splenectomy
Gharaibeh KI	2001	1	Left upper quadrant heaviness. On examination splenomegaly present	All investigations were suggestive of lower pole lesion in spleen	Laparoscopic splenectomy
Safioleas M et al.	1997	14	-	-	Open splenectomy
Aruna et al.	2021	1	Abdominal pain, fever and left upper quadrant swelling	CECT showed liver and spleen hydatid cyst with pericystic fluid, suggesting rupture	Enucleation of hydatid cyst with omentopexy with drainage of residual cavity

[Table/Fig-5]: Case reports/series of isolated splenic hydatid cyst [3,6,13-19].

CONCLUSION(S)

An adequate preoperative preparation and a meticulous surgical technique is required to prevent spillage of contents and to prevent anaphylaxis. Elective splenectomy should be done after completion of vaccination to prevent OPSI. In postoperative period, it is absolutely essential to continue antihelminthic therapy.

REFERENCES

[1] Gupta V, Kaira V, Sharma J, Sen R, Sangwaiya A. Primary hydatid cyst of the spleen: A rare entity. J Trop Dis. 2014;2(2):131.
[2] Park K. Park's Textbook of Preventive and Social Medicine. 21st Edition. Jabalpur: M/S Banarsidas Bhanot Publishers; 2011:798-806.
[3] Malik AA, ul Bari S. Human abdominal hydatidosis. Springer; 2019;73-77.
[4] Rasheed K, Zargar SA, Telvani AA. Hydatid cyst of spleen: A diagnostic challenge. N Am J Med Sci. 2013;5(1):10-20.
[5] Raichandani S, Chaturvedy KR, Gehlot R. Giant splenic cyst: A case report and consolidated review of literature with radiographic features. Indian J Case Rep. 2018;4(3):197-99.
[6] Kumar P, Hasan A, Kumar M, Singh V. Isolated hydatid cyst of spleen: A rare case with rare presentation. Int J Surg Case Rep. 2016;28:279-81. Doi: 10.1016/j.ijscr.2016.10.018. PMID: 27756032.
[7] Karabicak I, Yurtseven SS, Ozen N, Kesim M. Splenic hydatid cyst. Can J Surg. 2009;52(5):E209-10.
[8] Franquet T, Montes M, Lecumberri FJ, Esparza J, Bescos JM. Hydatid disease of the spleen: Imaging findings in nine patients. Am J Roentgenol. 1990;154(3):525-28. Doi: 10.2214/ajr.154.3.2106214. PMID: 2106214.
[9] Dachman AH, Ros PR, Murari PJ, Olmsted WW, Lichtenstein JE. Nonparasitic splenic cysts: A report of 52 cases with radiologic-pathologic correlation. Am J Roentgenol. 1986;147(3):537-42. Doi: 10.2214/ajr.147.3.537. PMID: 3526842.
[10] Garg M, Mangal A, Tak H, Singh DP, Soni A. Isolated large primary splenic hydatid cyst: A case report. Asian Pac J Trop Dis. 2015;5:S178-80.
[11] Santosh T, Patro MK, Bal AK, Behera B. Hydatid cyst at unusual locations: Report of two cases. Hum Pathol (NY). 2017;8:59-61. https://doi.org/10.1016/j.ehpc.2017.03.004. (https://www.sciencedirect.com/science/article/pii/S2214330016301377).
[12] Dehkordi AB, Sanei B, Yousefi M, Sharafi SM, Safarnezhad F, Jafari R, et al. Albendazole and treatment of hydatid cyst: Review of the literature. Infect Disord Drug Targets. 2019, 19(2):101-04.
[13] Vezakis A, Dellaportas D, Polymeneas G, Tasoulis MK, Chondrogiannis C, Melemeni A, et al. Two cases of primary splenic hydatid cyst in Greece. Korean J Parasitol. 2012;50(2):147-50.
[14] Hepgül G, Tıhan D, Kocael P, Doğan Y, Oztürk T, Cıhan A. Olgu Sunumu: Primer dalak kist hidatiği [Case report: Primary splenic hydatidosis]. Turkiye Parazitol Derg. 2010;34(3):184-86. Turkish. PMID: 20954121.
[15] Karakaya K. Primary hydatid cyst disease of the spleen; A rare entity: Report of two cases. Handle.net [Internet]. 2007. Available from: https://hdl.handle.net/20.500.12628/3159.
[16] Durgun V, Kapan S, Kapan M, Karabiçak I, Aydogan F, Goksoy E. Primary splenic hydatidosis. Dig Surg. 2003;20(1):38-41. Doi: 10.1159/000068864. PMID: 12637803.
[17] Gharaibeh KI. Laparoscopic excision of splenic hydatid cyst. Postgrad Med J. 2001;77(905):195-96. Doi: 10.1136/pmj.77.905.195. PMID: 11222831.
[18] Safioleas M, Misiakos E, Manti C. Surgical treatment for splenic hydatidosis. World J Surg. 1997;21(4):374-78.
[19] Aruna, Thapa G, Shah B. Hydatid cyst of spleen: A rare presentation. Indian J Pathol Oncol 2021;8(3):394-96.

PARTICULARS OF CONTRIBUTORS:
1. Junior Resident, Department of General Surgery, T.N.M.C. and B.Y.L. Ch. Nair Hospital, Mumbai, Maharashtra, India.
2. Junior Resident, Department of General Surgery, T.N.M.C. and B.Y.L. Ch. Nair Hospital, Mumbai, Maharashtra, India.
3. Assistant Professor, Department of General Surgery, T.N.M.C. and B.Y.L. Ch. Nair Hospital, Mumbai, Maharashtra, India.
4. Professor, Department of General Surgery, T.N.M.C. and B.Y.L. Ch. Nair Hospital, Mumbai, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Jayashri S Pandya,
B101, Gokul Monarch, Thakur Complex, Kandivali East,
Mumbai-400101, Maharashtra, India.
E-mail: smruti63@hotmail.com

PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Apr 26, 2022
- Manual Googling: Jun 28, 2022
- iThenticate Software: Sep 19, 2022 (17%)

ETYMOLOGY: Author Origin

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Mar 29, 2022**
Date of Peer Review: **May 26, 2022**
Date of Acceptance: **Jul 07, 2022**
Date of Publishing: **Jan 01, 2023**