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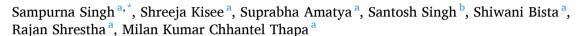
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Case Report

A case of giant primary splenic hydatid cyst: Case report



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ABSTRACT

Introduction: Primary splenic hydatidosis is a rare zoonotic disease, common in grazing regions of the world. Primary splenic hydatid cyst is rare and accounts for <2% of patients. Splenectomy is advocated in case of giant hydatid cyst locating in central and hilum area; however, spleen salvaging operations are done in superficial cyst, cysts localized to one pole of the spleen or cysts that are unresectable due to extensive adhesions and in children. Presentation of case: We present a case of 29-year male patient from rural areas presented with the complain of left hypochondriac pain and tenderness along with the history of farming and cattle raising. There was no other significant history.

Clinical findings and investigations: Physical examination showed mildly distended abdomen, mild tenderness over left hypochondrium. Routine laboratory investigations were sent. Ultrasound and CT scan of the abdomen showed giant splenic cyst measuring approx. $15 \times 14 \times 11$ cm along with daughter cysts. Postoperatively, cyst was sent for histopathological examination.

Intervention and outcome: Preoperatively, vaccinations against Pneumococcus, Hemophilus influenzae and Meningococcus along with Albendazole (15 mg/kg/day) was given 2 weeks before the planned operation. Total splenectomy was performed and diagnosis was confirmed by histopathological examination. Albendazole was continued for 2 more weeks. Patient remains asymptomatic thereafter and regularly followed up in OPD basis. Relevance and impact: Presence of isolated splenic cyst should raise suspicion for primary splenic hydatid cyst in endemic areas. Splenectomy versus spleen salvaging options should be analyzed and performed as per the indications. Splenic hydatid cyst is one of the rare clinical findings and there is very much high chance that it goes undiagnosed during the clinical practice. So, the main motive behind writing this article is to shed knowledge on basic approach to this splenic finding.

1. Introduction

1.1. Background and rationale

Berlot in 1790 was the first to describe splenic hydatidosis as an autopsy finding [1]. The hydatid disease is caused by Echinococcus granulosus and E multilocularis and the former is commonly seen in the great grazing regions of the world—particularly the Mediterranean region, Africa, South America, the Middle East, Australia, and New Zealand and is the most frequently encountered type of hydatid disease in humans [2]. Hydatid disease of spleen is a rare clinical condition as even in the endemic region the frequency is reported to be 0.5–4% of abdominal hydatid diseases [3]. Cases of splenic hydatid cyst (SHC) can be either primary (isolated to the spleen only) or secondary

(accompanied by hydatid cysts in other organs) [4]. Primary splenic involvement by hydatid cyst is rare and accounts for less than 2% of patients [5]. Secondary HD of the spleen can be caused by rupture of liver cysts with abdominal and pelvic dissemination [5].

Hydatid disease can develop in any organ, but the liver (60–70%) followed by the lung (30%) are the most frequent sites as they act as filters to the larvae in their journey in the blood stream [3,6]. Most of the larvae are trapped in the liver (first Lemman's filter) to form hepatic hydatid cysts; some larvae evade the hepatic microvascular barrier to reach the lungs (second Lemman's filter) forming pulmonary hydatid cysts and a small fraction of the released larvae bypass both filters and are distributed to different organs including the spleen, kidney, peritoneum, brain, bone, thyroid and mesentery [3,6,7]. Splenic echinococcosis may also arise by retrograde spread from the liver to the spleen via

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the hepatic portal and splenic veins in portal hypertension and the spleen can also be affected by rupture of a hepatic echinococcal cyst into the peritoneal cavity [7].

1.2. Guidelines: SCARE 2020 paper

This case report has been reported in line with the SCARE Criteria [8].

1.3. Patient information: demographics and presentation

A 29-year male farmer from the rural areas presented to OPD with the complaints of pain over left hypochondriac region for 2 weeks which was dull aching, continuous, non-radiating, aggravated on movement. There was history of cattle rearing including dogs and sheep in his native village in childhood.

1.4. Past medical and surgical history

No similar illness in the past. No known history of diabetes, hypertension, thyroid disorders and tuberculosis. There are no surgical interventions done in the past.

1.5. Drug and allergic history

No history of drug intake and no known allergy till date.

1.6. Family history

No similar illness in family members.

1.7. Social history

Does not smoke and does not consume alcohol.

1.8. Clinical findings

On examination, his vitals were within normal limits. Physical examination showed mildly distended abdomen, mild tenderness over left hypochondrium but no rebound tenderness and guarding. Other systemic examinations were normal.

1.9. Diagnostic assessment and interpretation

Laboratory investigations were done (Tables 1, 2). Ultrasonography of abdomen showed well defined splenic cyst

Table 1
Summary of preoperative laboratory test.

Laboratory test	Patient level	Unit	Reference range
Total bilirubin	1.4	Mg/dl	0.3–1.2
Direct bilirubin	0.3	Mg/dl	0.1-0.4
Alanine transaminase	71	IU/L	12-78
Aspartate transaminase	77	IU/L	15-37
Alkaline phosphatase	120	IU/L	46-116
Urea	12	Mg/dl	10-40
Creatinine	0.8	Mg/dl	0.3-1.2
Sodium	138	Mmol/L	135-146
Potassium	4.2	Mmol/L	3.5-5.2
Total leucocyte count	9410	/cumm	4000-11,000
Hemoglobin	16.5	Gm/dl	12-15
Blood sugar random	97	Mg/dl	70-140
Prothrombin time test	13.0		
International normalized ratio	0.92		
Blood grouping and Rh typing	A positive		
Serology	Non-reactive		
Urine routine	WBC: 0-1/HPF		
	RBC: Nil/HPF		

Table 2Summary of postoperative laboratory test.

Laboratory test	Patient level	Unit	Reference range
Amylase	744 (Drain fluid hemorrhagic sample)	IU/L	25–115
Urea	18	Mg/dl	10-40
Creatinine	0.7	Mg/dl	0.3-1.2
Sodium	134	Mmol/ L	135–146
Potassium	4.2	Mmol/ L	3.5–5.2
Total leucocyte count	17,220	/cumm	4000-11,000
Hemoglobin	13.8	Gm/dl	12-15

measuring $13.0\times11.8\times10.9$ cm in the anterosuperior aspect, smaller daughter cyst and irregular echogenic nodule is noted along the posterior wall suggestive of hydatid cyst (Figs. 1, 2). Other abdominal organs show normal findings.

Plain and IV contrast enhanced CT scan of abdomen and pelvis revealed enlarged spleen of 17cm along with well circumscribed cystic lesion measuring approx. $15 \times 14 \times 11$ cm (Figs. 3–5). Cystic lesion was hyperdense and had smooth margin; internal hyperdense septations and non-enhancing solid areas/debris were seen. The lesion was in the medial aspect of spleen abutting hilum and displacing hilar vessels, pancreas, splenic artery, stomach medially. Diaphragm and left kidney were abutted partially; minimal peri splenic collection, minimal left pleural effusion and minimal fluid in rectovesical pouch were present. Patient was diagnosed as case of primary splenic hydatid cyst and planned for elective splenectomy.

1.10. Intervention

Prophylactic therapy with Albendazole 15mg/kg/day was started immediately after the diagnosis and continued for 2 weeks before operation. Vaccination against meningococcus, Hemophilus influenza and pneumococcus were given 2 weeks before planned operation. After 2 weeks, patient was admitted and kept nil per oral for 8 hours prior to surgery. Left subcoastal incision was given. Intraoperatively, multiple cysts located within the enlarged spleen parenchyma crossing midline and extending up to the umbilicus was found. Cyst within the spleen was adhered to surrounding structures; adhered to the left hemidiaphragm superiorly, attached to the body and tail of pancreas posteriorly, medially was adhered to greater curvature of stomach and short gastric vessels. Medial and posterior aspect of the cyst was deroofed and splenectomy was done along with the removal of cyst (Figs. 6, 7). There was



Fig. 1. Ultrasonography of abdomen showing enlarged splenic cyst.



Fig. 2. Ultrasonography of abdomen showing enlarged splenic cyst with daughter cyst and echogenic nodule along its wall.



Fig. 3. CT abdomen showing enlarged cystic lesion in spleen with internal hyperdense septations and minimal peri splenic collection.



 ${\bf Fig.~4.}$ CT abdomen demonstrating enlarged spleen with well circumscribed cystic lesion.

no intraoperative spillage of cyst contents. Blood loss was minimal. Peritoneal cavity was washed with clean hypertonic saline and abdominal incisions were closed with sutures. Post-operative period was



 $\textbf{Fig. 5.} \ \ \textbf{CT} \ \ \text{abdomen showing enlarged cystic lesion in spleen with internal hyperdense septations.}$

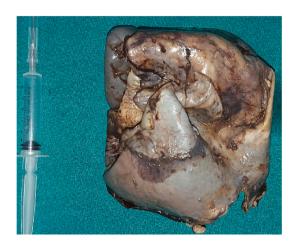


Fig. 6. Photo showing enlarged spleen after splenectomy.

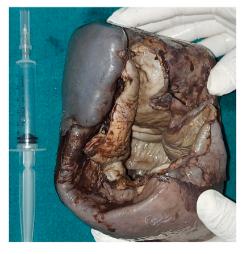


Fig. 7. Enlarged spleen with cavity after removal of cysts.

uneventful. The patient was relieved of his symptoms. Treatment with Albendazole was continued for two more weeks after surgery. Furthermore, the patient was managed with IV fluid Plasma-Lyte, Inj.

Piperacillin with Tazobactam, epidural analgesia with Bupivacaine and Morphine via epidural catheter, antipyretic, proton pump inhibitor and incentive spirometry. Histopathological examination of tissue sample shows laminated cuticular layer with focal presence of germinal layer which confirmed the diagnosis of hydatid cyst (Fig. 8).

1.11. Outcome

The patient had no complaints after the operation besides mild pain over the incision site. He is being followed up in OPD.

1.12. Intervention settings and name of clinician

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2. Discussion

Patients of splenic hydatid cyst remains asymptomatic for 5-20 years before the diagnosis is made [9]. Although the patient complaints are not so great as to interrupt their daily activities, hydatid disease is generally identified incidentally (up to 30%) during radiological investigations for some other reasons [1,10]. The symptom complex produced by splenic hydatid cyst are mainly due to mechanical displacement and pressure effect on the adjacent organs [9]. The main symptoms are abdominal discomfort or pain and palpable mass in the left upper quadrant [11]. The presentation of splenic hydatid disease can vary, including renal arterial compression and systemic hypertension or rupture of the splenic hydatid cyst to other organs [12]. There can be dyspepsia, constipation due to pressure on colon, dyspnea due to pushing up of the left diaphragm, fistula of the colon or perforation into the diaphragm or bronchial tree [1]. Rarely in some cases, lumbar pain is the only clinical presentation [12]. Echinococcal cysts of the spleen usually grow very slowly (approximately 2-3 cm per year) [9]. As the hydatic cyst increases in size it may lead to compression of the hilar vessels of the spleen, resulting in pericystic splenic atrophy; eventually the cyst may completely replace the splenic parenchyma and chronic pericystic inflammation may cause adhesion with adjacent organs or even fistulization between the cyst and nearby organs such as the stomach, pancreas, left colon, left kidney or bronchus [7].

Complications such as secondary infection, cyst rupture into the abdominal cavity and anaphylactic shock have been reported but seem mainly related to very large splenic cysts [13]. The traumatic or spontaneous rupture of a hydatid cyst may cause a life-threatening complication of systemic anaphylactic reaction [3]. Hypersplenism and

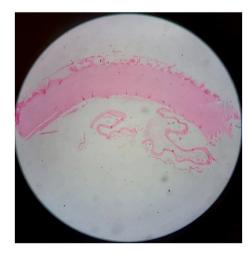


Fig. 8. Microscopic section of cyst showing laminated cuticular layer.

segmental portal hypertension due to hydatic cyst of the spleen is also reported [14].

Epidermoid cyst, pseudocyst, hematoma, abscess and cystic neoplasm have been considered as differential diagnoses of primary splenic hydatidosis [15].

The diagnosis is based on historical and geographic background, physical examination, serology and visualization of peripheral calcification or daughter cysts within the large cystic lesion or coexistent cystic lesions in the liver or other organs [9]. Routine laboratory investigations are of little help in diagnosis; eosinophilia is present in only 20–30% of the patients [14]. Ultrasonography is the first-line option for the screening of abdominal hydatidosis, its sensitivity for evaluating hydatid cyst is 90–95% and specificity is 93–100% [16]. Ultrasonographic classifications used in the liver hydatid cysts can also be used for splenic echinococcosis. An initial classification for cystic echinococcosis was proposed by Gharbi et al. (Table 3) [14].

Ultrasound and abdominopelvic CT scan allow a presumptive diagnosis showing cystic calcifications, daughter cysts or intra-cystic septa, hydatid sand, which are specific to echinococcal infestation [9,15], abdominal CT also helps in providing information regarding the cyst localization, neighborhood, type, and contents [10]; however, sonographic and CT findings of splenic cysts are not specific because other cystic lesions may present with similar appearances on US and CT [13]. Although CT scan is more sensitive than abdominal ultrasound, non-calcified benign cysts without daughter cysts cannot be differentiated per se from other benign cysts neither by computed tomography nor by ultrasound [17]. It has been found that MRI has some advantages in terms of detecting obscure cysts missed by USG and CT and of providing cystic pathological features, however, it is usually avoided because of its high costs [16]. Hydatidosis should be suspected in patients with splenic cystic lesions, particularly in endemic areas until proved otherwise and the diagnosis of splenic hydatidosis should be favored if daughter cysts are present within a large cystic lesion or if cystic lesions are observed in other organs such as the liver [1]. Diagnosis of splenic hydatidosis can be successfully established in more than 90% of all cases by USG and CT combined with immunological tests such as IHA, ELISA and immunoelectrophoresis [18]. ELISA has up to 84% sensitivity and it has been reported that the detection of specific IgG1 and IgG4 serum antibodies to echinococcosis can enhance the specificity of ELISA [9]. In general, cysts involving lungs, brain and spleen are associated with low serodiagnosis reactivity [19]. Once the diagnosis is made, systemic examination should be performed and other organs should be investigated for involvement [20].

Surgery is the mainstay of treatment for hydatid disease of the spleen [13]. Most surgeons advocate total splenectomy, especially for patients with large cysts located centrally or near the hilus, since it gives rise to minimal risk of recurrence [9]. Laparoscopy approach has been reported by some authors but has not yet been recommended as reference because of high risk of peritoneal spillage due to elevated intraabdominal pressure caused by pneumoperitoneum [11,16]. Laparoscopic splenectomy: the risk of rupture accompanied by anaphylaxis appears to be greater and besides, it is not applicable to big-sized cysts [19]. When compared with the open technique, the proven advantages of laparoscopy include good intraabdominal exposure, shorter hospital stay, less postoperative pain, and better cosmetic results [21].

Anaphylactic shock due to spontaneous or traumatic cyst rupture

Table 3Classification of hydatid cyst by Gharbi et al.

Type of cysts	Description	
I	Pure fluid collection	
II	Fluid collection with a split wall	
III	Fluid collection with septa	
IV	Hydatid cysts with heterogeneous echo patterns	
V	Hydatid cysts with reflecting thick walls	

during surgery is a rare and severe complication [22]. Splenectomy is associated with sepsis-related death in 1.9% in adults and 4% in children [23]. Although the risk of infection has been reported to be highest in the first 2 years after splenectomy affecting mainly the young, no age is exempt as overwhelming sepsis has been reported more than 50 years after splenectomy [6]. Surgical removal of the spleen results in loss of an important immune organ; there is reduced clearance of particulate extracellular and intracellular antigens including bacteria and malaria; impaired phagocytosis of opsonized and unopsonized bacteria and cells and decreased tuftsin and properdin levels [24]. Asplenic individuals are thus particularly vulnerable to sepsis caused by bacteria and protozoa and Babesiosis (transmitted by ticks in temperate and tropical countries) is almost completely confined to asplenic patients [6]. Therefore, all the patients should receive vaccinations against pneumococcal, Haemophilus influenzae and meningococcal infections at least 2 weeks before or after the splenectomy to reduce the risk of sepsis [4,6]. Splenectomy also increases the risk of developing various other diseases, including thromboembolic events, pulmonary hypertension, atherosclerosis and incomplete immunization with current vaccines [25]. Relapse rate after splenectomy varies between 0% and 12% [7,26].

Conservative surgery such as a partial splenectomy, cyst enucleation, or deroofing of the cyst with omentoplasty or external drainage have been performed for superficial cysts, cysts localized to one pole of the spleen or cysts that are unresectable due to extensive adhesions [18]. In a review of 333 published cases of splenic hydatidosis managed surgically, only 81 cases (24%) were treated by spleen sparing surgery [25]. Partial splenectomy has been recommended when the cysts are very large and located in the upper or lower pole [21]. The conservative procedures are not free of complications, hemorrhage after partial splenectomy or an infection of the residual cavity after deroofing the cysts can occur [18].

Although the preservation of the spleen is being increasingly advocated to prevent some complications associated with splenectomy, only the complete removal of the diseased organ rids the patient of a potentially infected cavity which carries the risk of recurrence [13]. However, a study of 19 patients with splenic hydatid cysts found no significant difference between the splenectomy and spleen preserving groups concerning median hospital stay and postoperative complication rate [7]. Since the type of surgery (splenectomy or spleen-sparing procedures) does not appear to influence patient outcome, the less-extensive approaches may be preferred according to their relatively lower risk and long-term effects on the patient [4].

Use of antihelmintic agents (albendazole, mebendazole and praziquantel) as the only treatment modality remains controversial whereas it is considered useful as adjuvant therapy before or after surgery [25]. The most frequently administered agents are albendazole (10–15 mg/kg/day), mebendazole (40–50 mg/kg/day), and praziquantel (25 mg/kg/day) [4]. Medical prophylaxis with albendazole or mebendazole should be given to patients perioperatively as well as during the first 6 months after surgery to prevent the recurrence [27].

3. Conclusion

Hydatid cyst in liver and lung is common finding; however splenic hydatid cyst is a rare clinical entity even in endemic areas. Hydatid cyst is common in grazing areas of the world. Its presentation can widely vary from being asymptomatic in some patients to presenting with symptoms like left upper quadrant pain and abdominal distension in others. Ultrasonography and CT scan of the abdomen showing cystic calcifications, intra cystic septa or daughter cysts along with hydatid sand combined with serologic testing like ELISA confirms the diagnosis of splenic hydatid cyst. Primary splenic hydatid cyst should be kept as differential in case of cystic lesions in spleen in endemic areas until proven otherwise. Total splenectomy is the preferred choice of treatment in giant, central splenic hydatid cyst involving hilar area, however partial splenectomy and other spleen preserving surgery are advocated

in case of superficial cysts, cysts located in one pole, in case of extensive adhesions and in children. Perioperative vaccinations and albendazole therapy should be recommended to prevent the risk of overwhelming post splenectomy infections and recurrences.

Ethical approval

This type of study does not require any ethical approval by our institution.

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This study did not receive any sources of funding.

Author contribution

Sampurna Singh, Internship at Department of Surgery, Kist Medical College, Kathmandu: He has conceptualized the study, collected images, analyse data, wrote the case and done all the discussion part. Shreeja Kisee, Internship at Department of surgery, Kist Medical College, Kathmandu: She has collected data, images, wrote history and examination portion. Suprabha Amatya, Internship at Department of Surgery, Kist Medical College, Kathmandu: She has collected data and images, helped in writing. Santosh Singh, Department of surgery, Gandaki Medical College, Pokhara: he has helped in literature review, done discussion portion and edited the article. Shiwani Bista, Internship at Department of Surgery, Kist Medical College, Kathmandu: Editor of the article. Rajan Shrestha, Internship at Department of Surgery, Kist Medical College, Kathmandu: He had reviewed the article and gives valuable suggestions. Milan Kumar Chhantel Thapa, Internship at Department of Surgery, Kist Medical College, Kathmandu: Editor of the article.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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