

## CASE REPORT

# A giant splenic hydatid cyst: Why calcified cysts should not be considered as a dead cyst

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## Abstract

Our case report showed that peripheral wall calcification of the hydatid cyst does not mean inactivation of the cyst and calcification of cyst wall may occur in all stages of disease.

## KEYWORDS

calcification of cyst, infectious diseases, splenic hydatid cyst

## 1 | INTRODUCTION

We aimed to present a splenic hydatid cyst diagnosed coincidentally in a patient with dull abdominal pain, and despite calcification of cyst wall in imaging findings, postoperative gross and pathological evaluations reported active hydatid cyst. Our case showed that calcification of the cyst is not dependable for calculating of activation.

Hydatid disease occurs in 2%-6% of endemic populations, and its annual incidence in Europe is rising in some regions.<sup>1-4</sup> Hepatic tissue along with lungs is the most commonly affected organ in humans.<sup>5</sup> The occurrence of primary splenic hydatid cyst (SHC) is not common even in endemic regions. The primary SHC involves splenic tissue only while the secondary form is diagnosed with the existence of hydatid

cysts in multiple organs.<sup>6</sup> The progress of hydatid cysts is 0.3-2.0 cm per year, and due to its slow expansion, most of the patients have nonspecific symptoms or even asymptomatic. Due to nonspecific clinical symptoms and rarity in existence, diagnosis of SHC is somewhat complicated and difficult.<sup>7</sup> Various paraclinical and imaging approaches such as serological tests, ultrasonography (US), and computerized tomography (CT) scans are useful for SHC diagnosis.<sup>8</sup> The selection of effective approaches for managing SHC correlates with the size and location of the cysts and stage of the diseases. Surgical intervention like splenectomy is the mainstay of treatment.<sup>9</sup> We aimed to describe an SHC case with nonspecific symptoms diagnosed incidentally in a patient with dull abdominal pain for a long time. The current case report was written in accordance with the SCARE guideline.

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## 2 | CASE PRESENTATION

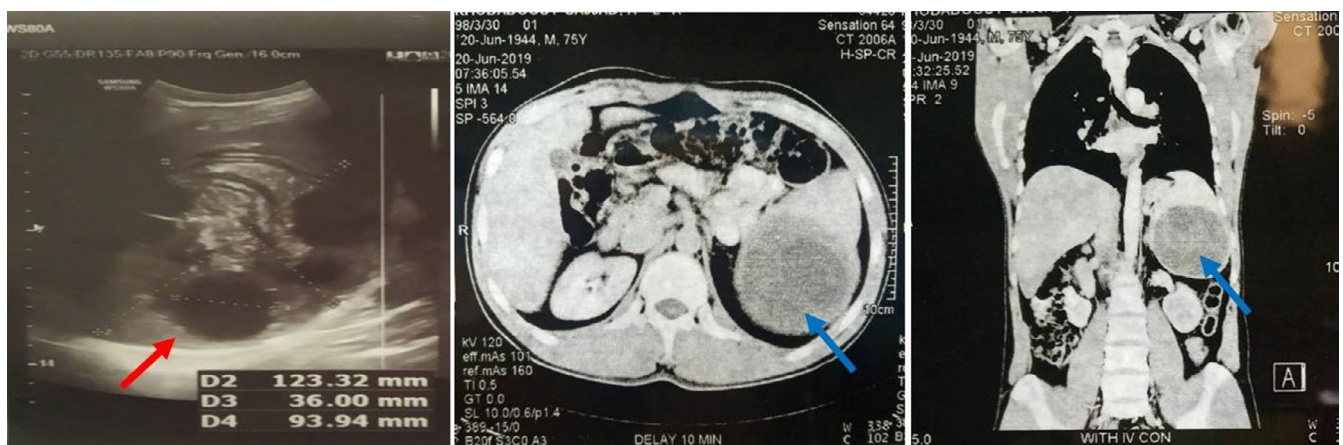
A 29-year-old male suffering from chronic intermittent pain of abdomen, especially in left flank and LUQ over the past 3 years, came to our hospital. In an exhaustive history taking, the patient had a dull background aching abdominal pain which has no clear relationship with eating and defecation. Despite any clear relationship between the pain and eating or defecation, he was treated with irritable bowel disease suspicion. Because of maintenance of the pain, while the patient represents a vague history of urinary tract symptoms such as dysuria or frequent urination before, with or after the pain, he was referred to an urologist with suspicion of uroliths. No signs of nephrolithiasis were found in examination and kidney, ureter, and bladder X-ray. In the US imaging, kidneys were normal while an enlarged pampiniform plexus veins in both scrotums (Grade II varicocele) found which seemed irrelevant to the pain. Regarding the patient's complaint to the existence of chronic pain on the left side of the abdomen, the radiologist assessed the left upper abdominal quadrant despite the main physician's focus on urinary tract disorders. A calcified hyperco-heterogeneous internal cyst was observed with an approximate size of 110 × 93 mm in the splenic parenchyma suitable with stage 5 of Garbi's classification of cysts<sup>10</sup> (Figure 1). The patient was subjected to contrast-enhanced computerized tomography (CECT) scan. CT demonstrated relative splenomegaly and evidence of well-defined complex cystic lesion of the approximate size of 106 × 92 × 93 mm in the splenic parenchyma. The cyst margin was calcified while internal fluid with hyperdense membranes suggested the existence of a typical hydatid cyst (Figure 1). The involvement of other organs was not shown. The patient was subjected to surgical approach and given 10 mg/kg/day albendazole for 3 days preoperatively. The cyst was centrally located close to the hilum. Total splenectomy performed because of

the location and size of the cyst. Gross and histopathological examinations reported active “hydatid cyst” as shown in Figure 2A and 2B. Different layers of the cyst were observed in microscopic examination. No postoperative sepsis or infectious complications were seen. After splenectomy, the patient experienced 7 days of hospital stay and took 10 mg/kg/d albendazole for 21 days orally. The patient has been vaccinated preoperatively with a single dose of pneumococcal polyvalent vaccine. Patient's preoperative and postoperative paraclinical data showed in Tables 1 and 2.

He was followed up every 3 months for 1 year, and ultrasound imaging was performed every 6 months to evaluate the probability of recurrence. No postoperative recurrence was recognized in the periodic follow-up.

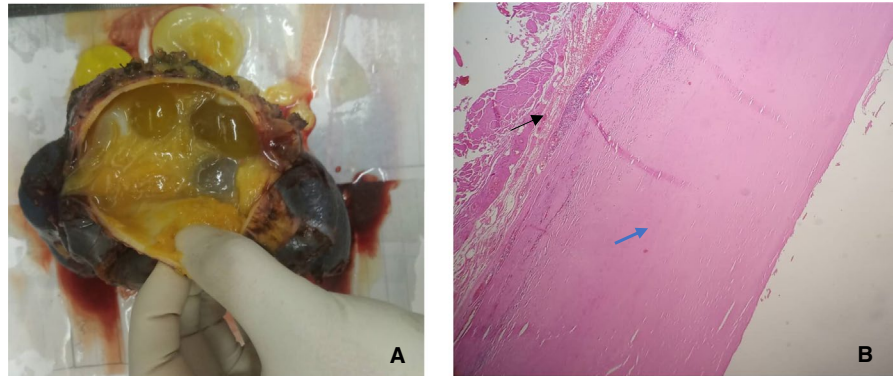
## 3 | DISCUSSION AND CONCLUSIONS

Hydatid disease is known as an important public health problem in the endemic regions, including Asia, Mediterranean, Middle East, South America, New Zealand, and Australia, and has been detected in many organs but commonly occur in liver (55%-60%) and lung (30%) tissues,<sup>11</sup> and other organs with less prevalent including kidney (2.5%), heart (2.5%), bones (2%), muscles (1%), spleen (1.5%), and brain (0.5%).<sup>12</sup> Clinical manifestations depend on the size of the cyst and type of organ involved. The occurrence of SHC is usually asymptomatic, and patients are often diagnosed coincidentally (23.8%-28.5%).<sup>13,14</sup> SHC symptoms include a dull aching pain in the LUQ (14.2%-100%) with or without a palpable mass (9%-75%), nausea and vomiting, heartburn, unintentional weight loss, dyspepsia, dyspnea, infection, constipation, and rupture or fistulization extended to the colon.<sup>11,13-15</sup> In our case, the patient was suffering from a left



**FIGURE 1** Ultrasonography diagrammatic and CECT scan splenic findings. The red arrow shows the hyperco-heterogeneous cyst in ultrasonography imaging, and the blue arrows show a complex cystic lesion in CECT with calcified margin and internal fluid with hyperdense membranes

**FIGURE 2** Illustration of splenic tissue with hydatid cyst composed of caseous material and numerous daughter cysts (A). Histological analysis of different layers of hydatid cyst (B). Hydatid cyst wall/ laminated cyst wall (blue arrow) and inner germinal layer (black arrow)



**TABLE 1** Preoperative paraclinical data

A. Hematology			
WBC	4000	Segment	2390
		Lymphocyte	1280
		Mixed	330
RBC	$4.55 \times 10^6$	Hb	12.2 mg/dL
		Hct	38.1%
		MCV	84 fL
		MCH	27 Pgm
		MCHC	32%
		RDW	14%
Platelet	195 000	PDW	11.9 fL
		MPV	9.7 fL
		P-LCR	23.7%
B. Biochemistry			
FBS		117	
Urea		41	
Creatinine		1.08	
Serum Na		141	
Potassium		3.8	
AST		13	
ALT		9	
Alk P		197	
Bilirubin		0.8	
Amylase		39	
Lipase		27	

Note: Patient's preoperative paraclinical data: A. Hematology and B. Biochemistry

**TABLE 2** Postoperative paraclinical data

A. Hematology			
WBC	11 000	Segment	80.3%
		Lymphocyte	9.6%
		Mixed	10.1%
RBC	$4.91 \times 10^6$	Hb	14.7 mg/dL
		Hct	41%
		MCV	84 fL
		MCH	30 Pgm
		MCHC	36%
		RDW	13.8%
Platelet	277,000	PDW	13.5 fL
		MPV	10.5 fL
		P-LCR	29.9%
B. Biochemistry			
FBS		136	
Urea		28	
Creatinine		1.23	
Serum Na		141	
Potassium		3.7	
AST		19	
ALT		13	
Alk P		205	
Bilirubin		0.9	
Amylase		47	
Lipase		30	

Note: Patient's postoperative paraclinical data: A. Hematology and B. Biochemistry

flank and LUQ dull pain with an unclear history of urinary tract symptoms.

US and CT assessments are the most valued diagnostic tools for detecting hydatid cysts in suspected patients and may present as a hypoechoic and/or anechoic cysts with hyperechoic marginal calcification, and/or daughter cysts. Besides, floating membranes may be detectable in the US examination.<sup>1,7</sup> Imaging findings of SHC containing

calcification of the cyst wall, presence of daughter cysts, cystic membranes, septa or hydatid sand, and presence of calcification and/or daughter cysts could rule out other cystic lesions of spleen. MRI studies a cystic lesion represented with low-signal intensity border on T2-weighted images.<sup>7,16,17</sup> Radiological studies are useful for indicating the location of the cyst, its impingement on the neighboring tissues, and planning the ideal treatment approach.<sup>18</sup>

Previous studies noted the effectiveness of ELISA and Casoni in the diagnosis of SHC.<sup>1,4</sup>

In this case report, the patient was also diagnosed by US examination and CT findings. No serologic evaluation was carried out. CT demonstrated evidence of well-defined cystic lesion in the splenic parenchyma with a calcified margin and internal fluid with hyperdense membranes suggested the existence of a typical hydatid cyst. Commonly, calcification of the cyst is interpreted in favor of cyst death, and in the literature review, there is a few data support to the presence of vesicles or protoscolices in calcified cysts and calcification of the cyst wall is accepted as an important sign of cyst death.<sup>19</sup> Hosch et al reported that peripheral wall calcification of the hydatid cyst is not restricted to the inactive cyst, but may occur in all stages in up to 50% of cysts.<sup>20</sup> In some studies, it has been demonstrated that calcification of the cyst wall does not mean cyst death.<sup>21</sup> Erzurumlu et al reported that live protoscolices were detected in the parasitological examination of a calcified hydatid cyst.<sup>22</sup> As in our case and mentioned studies, calcified cysts may probably contain dead vesicles that coexist with live ones. Consequently, we recommended subsequent follow-up every six months by imaging methods in cases with calcified cyst wall which are not candidates for a surgical approach. The cyst's viability could be indicated based on morphological changes.

Based on our previous experiences and reviewing the lectures, it seems that because of nonspecific presentation of SHC, longtime dull abdominal pain especially in LUQ should be considered as SHC as a differential diagnosis in an endemic area.<sup>11,13-15</sup> It seems that the symptom complex induced by SHC is generally due to mechanical displacement and pressure effect on the adjacent organs, and as seen in our case, chronic pericystic inflammation may trigger inflammatory process with adjacent and nearby organs such as stomach, pancreas, left colon, and left kidney.<sup>13</sup> Also, traumatic or spontaneous cyst rupture or during surgery can lead to anaphylactic shock in some patients and cause death if the diagnosis is not recognized, while appropriate treatment could not immediately performed or anaphylaxis be refractory to treatment.<sup>23</sup> Usually, secondary SHC occurs as a result of intraperitoneal spread following hepatic hydatid cyst rupture or systemic dissemination of echinococcal cysts.<sup>24</sup> The probable hypothesis of isolated hydatid cysts of spleen includes arterial and venous routes; thus, the arterial pathway occurs after passing through the liver as the first filter and through lung as the second filter. In the venous pathway, the infectious agent bypasses the liver and lung through portal circulation.<sup>25</sup> Early diagnosis of SHC could prevent further dissemination, seeding, or subsequent anaphylactic shock.<sup>12</sup>

Treatment is mainly based on surgical approaches and options depend on the individual patient, level of disease,

and surgeon's expertise. Surgical approaches contain total splenectomy, partial splenectomy, cyst unroofing, and enucleation with omentoplasty.<sup>26,27</sup> Spleen-preserving approaches are considered technically simpler and safer compared with the splenectomy method. However, residual cavity and recurrence may occur more in the methods.<sup>13,18,26</sup> In patients with large cysts located centrally or close the hilus, total splenectomy is recommended, since it makes the minimal risk of recurrence and rupture of cyst during surgery.<sup>28,29</sup> Ran et al performed total splenectomy in cysts larger than 10 cm in size and cysts located in the central spleen parenchyma.<sup>18</sup> In our study, the total splenectomy procedure via laparotomy was used due to the large size and centrally location of the cyst and received 10 mg/kg/d albendazole for 21 days orally while previously medical treatment with albendazole approved after surgical treatment to reduce the incidence of recurrence.<sup>30</sup> Laparoscopic surgery was not used considering the size and location of the cyst and higher risk of rupture.

Our report highlights the incidence of rarely isolated SHC with unspecified presentation and calcified margin in imaging findings which seem to be inactive while after surgery and during pathological gross and microscopic assessment cleared that the cyst was live and may cause life-threatening conditions.

In conclusion, our report emphasizes that calcification of the hydatid cyst is not dependable for calculating the activity of the cyst. The viability of cyst should be assessed by the evaluation of the content and behavior of cyst during follow-up instead of consideration of wall calcification in imaging findings.

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Published with written consent of the patient.

## CONFLICT OF INTEREST

None declared.

## AUTHOR CONTRIBUTIONS

H. A., A. R., and RR: were major contributors in writing the manuscript. H. A. and SH: were the attending physicians and operators of this patient. RR: confirmed the pathological finding. All authors read and approved the final manuscript.

## ETHICS APPROVAL AND CONSENT TO PARTICIPATE

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

## DATA AVAILABILITY STATEMENT

The datasets used during the current study are available from the corresponding author on reasonable request.

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