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Large expanding splenic pseudocyst: A case report and review of literature

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ARTICLE INFO	A B S T R A C T
<i>Keywords:</i> Splenic pseudocyst Non-parasitic splenic cyst Splenectomy Case report	Introduction and importance: Splenic pseudocysts are extremely uncommon. Most of these cysts are asymptomatic and may result from previous blunt abdominal trauma. We report an interesting uncommon case of large splenic pseudocyst without history of previous abdominal trauma. <i>Case presentation:</i> A 56 year old male patient, presented with symptoms of pain in the left side of middle back and discomfort in the left hypochondrium for few months. His physical examination was unremarkable. The abdominal Ultrasound and contrast-enhanced Computed tomography showed a large splenic cyst occupying most of the splenic parenchyma. <i>Echinococcus multilocularis</i> antibody test was negative. The differential diagnosis of this case included non-parasitic splenic cysts. The patient underwent elective exploratory laparoscopy which was converted to laparotomy with total splenectomy. Histopathological examination of the specimen revealed a splenic pseudocyst. <i>Clinical discussion:</i> The splenic cyst in this case was symptomatic due to its large size. It was hard to elicit an etiology as there was no history of abdominal trauma, infection, or degenerative disease. The main factors in selecting either conservative or radical surgical approach for such cases are the cyst location, cyst size, and the residual splenic parenchyma. <i>Conclusion:</i> The goal of splenic pseudocysts treatment is to relieve symptoms and avoid complications. Partial splenectomy is the recommended procedure when the size and location of the cyst allow preservation of at least 25% of splenic parenchyma. Otherwise, Total splenectomy is unavoidable.

1. Introduction

Splenic cysts are extremely uncommon, and only occur in about 0.07% of the population [1]. There are around 800 cases reported in the world literature, and one of their rarest types is the splenic pseudocyst [2–6]. The most common cause of splenic pseudocysts is previous blunt abdominal trauma in 75% of cases [2,7–9]. Other possible causes include infection, infarction, and degenerative diseases.

To be properly treated, it is important to distinguish splenic pseudocysts from other types of splenic cysts. Most splenic cysts (30–60%) are asymptomatic and are diagnosed incidentally during imaging studies [10,11]. The treatment options include cyst aspiration, fenestration and drainage, partial splenectomy, or total splenectomy. We report an interesting case of large expanding splenic pseudocyst with no previous abdominal trauma.

This case report has been reported in line with the SCARE Criteria [12].

2. Case presentation

A 56 year old western male patient was referred from a medical center to Al Ahli Hospital Urology clinic for evaluation of left renal cyst that appeared on abdominal imaging.

The patient was complaining of pain in the left side of middle back and discomfort in the left hypochondrium for few months. This complaint was not accompanied by nausea, vomiting, chest pain, cough, fever, or urinary symptoms. He had no history of abdominal trauma in the recent past.

The patient was married, non-smoker, non-alcoholic, and had an office job. He had a history of hypothyroidism, hypertension, and dyslipidemia, and was on oral medical treatment including: Levothyroxine, Atorvastatin, and Valsartan/Hydrochlorothiazide. He had no history of previous surgery, and no known drug allergy. His family history did not include any relevant genetic information, or psychosocial history.

The physical examination was unremarkable. The abdomen was soft

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and lax with no tenderness, palpable masses, or jaundice.

X-ray of lumbosacral spine showed mild scoliosis of lumbar spine, and reduced disc space at L1-L2, L2-L3, and L5-S1.

Contrast-enhanced computed tomography of abdomen showed a well-defined cystic lesion with diffuse wall calcification at upper pole of the spleen measuring $98 \times 95 \times 90$ mm, with no septa/soft tissue nodules. The stomach was slightly displaced to the right side. There was a large simple renal cortical cyst at posteromedial aspect of the upper pole of the left kidney measuring $88 \times 78 \times 72$ mm, Bosniak category I (Fig. 1).

Abdominal ultrasonography confirmed the presence of a large splenic cyst with internal echoes.

The urologist concluded that the left renal cystic lesion did not need any surgical intervention. Then the patient was referred to the general surgery clinic on the same day for evaluation.

Hematological and biochemical tests were within normal limits.

Echinococcus multilocularis antibody test was negative (titer <1:16), Thus the Echinococcosis was excluded from the differential diagnosis.

Laparoscopic surgical treatment of the symptomatic splenic cyst has been recommended, with possible conversion to laparotomy and splenectomy.

The patient was given preoperative vaccination in line with recommendations of Center of Disease Control and prevention (CDC), due to the predictive possibility of a subsequent splenectomy.

He was kept fasting for 8 h, and a dose of prophylactic intravenous



Fig. 1. CT scan of the abdomen, Axial view showing a large splenic cyst with calcified wall, occupying most of the splenic tissue.

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antibiotic was given before the surgery. Then he underwent an elective exploratory laparoscopy under general anesthesia in a supine position. Intraoperatively, a large splenic cystic lesion of hard calcified wall was found occupying more than 75% of the spleen parenchyma (the upper pole, middle part, and significant part of the splenic hilum). The cyst was firmly adherent to the diaphragm and left lobe of the liver, and covered with omental adhesions. There was difficulty in releasing these adhesions due to the hard calcified wall of the cyst. The procedure was converted to laparotomy through a left subcostal incision. Total Splenectomy was done with no intraoperative complications (Fig. 2).

The procedure was performed by senior specialist general surgeon and senior consultant general surgeon, in a highly equipped operation room in Al Ahli Hospital, Doha, Qatar.

The patient had an uneventful postoperative recovery course. He was discharged from the hospital on the third postoperative day (within the expected timeframe), and instructed to avoid heavy lifting for few months, and to complete the post-splenectomy vaccination protocol.

Histopathologically, the spleen weighed 680 g and had size of 13 \times



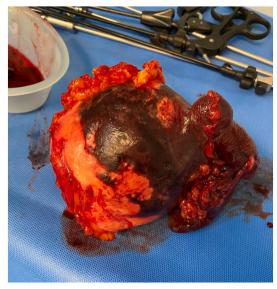


Fig. 2. Intraoperative images of the cyst lesion occupying most of the splenic tissue.

 10×7.5 cm. There was a splenic cyst measuring 9.7 cm. The cyst contained thick yellowish-white necrotic material and had a thick fibrotic wall. The wall thickness was up to 1.2 cm with areas of calcifications. Microscopic examination showed that the cyst wall was composed of dense fibrous tissue without an epithelial lining and showing extensive foci of calcification. The adjacent splenic tissue was unremarkable. The appearance was consistent with splenic pseudocyst (Figs. 3 & 4).

Postoperatively, the patient was followed up regularly in the surgery clinic for 8 months, and neither symptoms nor related complaints recurred. He was compliant with the instructions, enjoying a good quality of life, and grateful for the achieved clinical outcome which was satisfactory and similar to his expectations.

3. Discussion

Splenic cysts refer to cystic lesions of the spleen. The reported incidence of splenic cysts is only around 800 cases globally [2–6]. Andral described the first non-parasitic cyst of the spleen in 1829 [13,14]. Fowler published a collective review in 1953 including 265 cases of nonparasitic splenic cysts [15]. A series of 42,327 autopsies over a 25-year period was reported by Robbins in 1978, revealing only 32 patients with splenic cyst [1]. The first splenectomy for splenic cyst was in 1867 by Pean [14]. The first classification of splenic cysts included two types: true cysts (with lining epithelium), and false cysts (without lining epithelium) [10,15–17]. Martin divided the splenic cysts as following: type I cysts are primary (true) cysts with an epithelial lining (either parasitic or nonparasitic in nature), and type II cysts are secondary (false) cysts without an epithelial lining [16]. Later on the splenic cysts were divided into two types: parasitic and nonparasitic cysts, and nonparasitic ones were classified into primary (epithelial/true) and secondary (false/pseudo) types [17]. Parasitic cysts account for more than two thirds of the cases in the endemic areas. The most well-known etiology of these cysts is Echinococcus granulosus [14]. Recently, a pathological classification divided nonparasitic cysts into congenital, traumatic, neoplastic, and degenerative types [10,17].

Splenic pseudocysts are four times more common than true cysts. They constitute 70–80% of all the cases, particularly affecting women, children, and young adults [2,14,18,19]. The etiology of splenic pseudocysts refers to the following: resolved hematoma due to a previous blunt injury, infections, infarction, or degenerative diseases [2,18,20–22].

There are some rare conditions as following:

- Intrasplenic pancreatic pseudocysts [23].
- Subcapsular or intraparenchymal splenic hematomas in the course of acute or chronic pancreatitis [24].

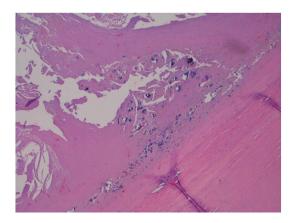
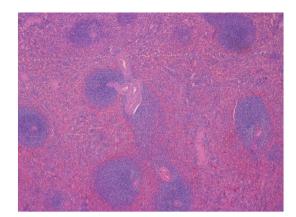
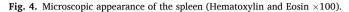


Fig. 3. Microscopic appearance of cyst wall (Hematoxylin and Eosin ×100).





- Spontaneous subcapsular hematomas associated with infectious mononucleosis [25], cytomegalovirus infection [26], cocaine use [27], or fiberoptic colonoscopy [28].

Most splenic pseudocysts (30–60%) are asymptomatic [6]. The majority of reported cases are diagnosed accidently due to the lack of specific clinical symptoms in the early stages [22], but oversized cysts tend to be symptomatic [10,22]. The symptoms correlate with the size and location of the cyst [29]. The most common symptom is atypical pain and heaviness in the left hypochondrium due to distension of the splenic capsule or space-occupying mechanisms within the abdominal cavity. Other symptoms include left shoulder pain, chest pain, nausea, vomiting, dysphagia, dyspnea, persistent cough, or may appear as a palpable mass [10,11,30–32].

Serious complications have been reported in large splenic pseudocysts, attributed to sudden increase in the cyst size due to intra-cystic bleeding, secondary infection, and spontaneous intraperitoneal rupture [2,3,6,33]. Cowles found a complication rate of 5.2%, namely rupture and infection in 191 total cases [34].

The radiological diagnostic tools include: Ultrasonography test (USG), Contrast-enhanced computed tomography (CECT), Magnetic resonance imaging (MRI), and Magnetic resonance angiogram (MRA) [14,18]. Splenic pseudocysts may be found incidentally during an abdominal ultrasound or may be detected due to calcification on radiographs [32]. USG is useful to determine the nature of the cyst content, presence of calcification, presence of septa, and regularity of the cystic wall [35]. CECT helps to evaluate the cyst morphology, cystic fluid, and the relationship between the cyst and the surrounding organs [22,35]. However, the above techniques cannot distinguish between true cysts and pseudocysts. Therefore, the gold standard for clinical diagnosis of splenic cysts remains the histopathology.

The majority of splenic pseudocysts are unilocular and have a smooth wall. Microscopic findings consist of fibrous wall tissue without an epithelial lining [10,30,36].

Splenic pseudocysts larger than 5 cm usually require surgical treatment [37,38]. Cysts with dimensions greater than 15 cm are called giant cysts [10,18,30,39]. The traditional approach to splenic pseudocysts larger than 5 cm has been total splenectomy, but the current approach has become more conservative including partial splenectomy, aspiration, deroofing, marsupialisation, decapsulation, and cystectomy [5,18,39–41]. Changes of therapeutic approach resulted from increased knowledge about the importance of the reticuloendothelial and hematopoietic role of the spleen, and overwhelming life-threatening septicemia, especially in children who underwent splenectomy.

Laparoscopic deroofing and aspiration have been reported with a 20-40% recurrence rate [33,42]. Schier reported the laparoscopic deroofing to have a recurrence rate of 64% within an average of 1 year, despite all the procedures used to treat residual cavity (omentum package, argon destruction) [43]. Percutaneous drainage with or without infusion of sclerotic substances has been associated with high recurrence rate and does not result in long term control [44,45]. Partial splenectomy appears to be an appropriate procedure if at least 25% of the splenic parenchyma is preserved [46,47]. It is recommended to be performed by a laparoscopic approach, if the cyst is located in the poles of the spleen [48]. Total splenectomy is recommended only for huge cysts, cysts covered by the splenic parenchyma, or cysts located near the hilum [49,50]. Certain characteristics of splenic cysts including hilar location, large size with near complete replacement of parenchyma, associated hypersplenism, and doubtful diagnosis are the few important situations where total splenectomy may not be avoidable [5,40,51].

Our patient developed symptoms due to the large size of the splenic cyst, and his medical history did not reveal any significant information leading to its etiology. Diagnosis was made based on abdominal USG and CECT findings. The hematological, biochemical, and serological tests were within normal limits, indicating a non-parasitic nature of the splenic cyst. We performed a total splenectomy due to the location and size of the cyst, in addition to the insufficient residual splenic parenchyma. Histopathological examination confirmed the diagnosis of large splenic pseudocyst.

4. Conclusion

Splenic pseudocysts are uncommon pathology of benign nature, often secondary to resorption of a hematoma. Sometimes it is hard to elicit a clear etiology of these cystic lesions. The goal of splenic pseudocysts treatment is to relieve symptoms and avoid complications. When they are symptomatic, a surgical intervention is recommended based on cyst size and location, and complications such as rupture, hemorrhage, and infection. Definitive diagnosis is possible only by histopathological examination. Partial splenectomy is the recommended procedure when the size and location of the cyst allow at least 25% of splenic parenchyma to be preserved. Otherwise, total splenectomy is unavoidable.

Ethical approval

This case report was approved by the ethics committee, Al Ahli Hospital, Doha, Qatar. A copy of the approval letter is available for review by the Editor-in-Chief of this journal on request.

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CRediT authorship contribution statement

Bakhos Alhaddad: study concept and design, data collection and analysis, writing the paper.

Abdul Azim Hussain: study concept and design, supervision, final approval of the paper.

Haitham Al-Rawi: data analysis and interpretation.

Rafif Mahmood Al Saady: data analysis and interpretation, writing the paper.

Guarantor

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Research registration

None.

Consent

Verbal and written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Case presentation disclosure

I disclose that this case has not been presented at a conference or regional meeting.

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Declaration of competing interest

The authors declare no potential conflict of interest related to this manuscript.

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