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Oversized pseudocysts of the spleen: Report of two cases Optimal management of oversized pseudocysts of the spleen☆

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ABSTRACT

INTRODUCTION: Pseudocysts of the spleen are usually asymptomatic and associated with a history of trauma, infection or infarction. In this report, we present two uncommon cases of solitary, oversized pseudocysts of the spleen.

PRESENTATION OF CASE: Two patients (cases A and B), with symptoms of abdominal pain, were investigated. The laboratory and ultrasound examination confirmed the diagnosis of a large, non-parasitic splenic cyst in both cases. Computed tomography described an oversized pseudocyst occupying almost the entire splenic parenchyma in both cases and in patient A, the cyst was located in the splenic hilum. The medical history revealed a previous abdominal injury only in case A. The two patients underwent an open total splenectomy. The pathology examination verified the diagnosis of a non-parasitic splenic pseudocyst.

DISCUSSION: Both patients presented with symptoms, in contrast to the majority of patients with splenic cysts. The medical history of patients with splenic pseudocysts does not always reveal the cause of the pseudocyst formation. Any type of spleen-sparing procedure is not easy to perform in cases of surgical and anatomical difficulty, because of recurrence and the risk of intractable bleeding from the spleen.

CONCLUSION: Partial splenectomy is the recommended method for parenchymal preservation, but total splenectomy is preferred when the splenic cyst is oversized or cannot be excised with safety.

1. Introduction

Cystic lesions of the spleen include benign cysts, neoplasms and abcesses. Benign splenic cysts are an everyday surgical entity. The classification has been introduced by Martin et al., about 53 years ago.[1] Type I cysts (true, primary cysts) can be parasitic (hydatosis), caused mostly by the parasite *Echinococcus granulosus*, or non-parasitic. They are characterized by an epithelial cover or an endothelial cover of their inner wall. Type II cysts (secondary cysts) are false cysts (pseudocysts), which do not have an epithelial lining. Splenic pseudocysts (SPs) originate usually from a past injury, infection or infarction.[2]

Regarding epidemiology, parasitic cysts are usually found in underdeveloped countries of Africa or South America, whereas non-parasitic cysts are commonly found in developed countries of Europe and North America, possibly due to their etiology.[3] The prevalence and diagnosis of splenic cysts have increased recently, mostly due to the increase in the use of computed tomography (CT) and due to the successful conservative management of splenic injuries, infarctions or infections.[3,4] The true incidence of splenic cysts is unknown, but blunt trauma of the abdomen is considered the commonest cause of later formation of a secondary splenic cyst (pseudocyst).[4] Almost 75% of all SPs are post-traumatic.[4]

Concerning the clinical presentation, most of the cysts (30–60%) are asymptomatic and diagnosed incidentally.[3,5] The SPs are a type of cysts, which cannot be clinically distinguished from other types. The cysts do not produce symptoms, unless they are oversized.[2,3] Most of the SPs are unilocular (80%) and they vary in size (1–16 cm). Large cysts may cause atypical pain and heaviness in the left hypochondriac region, due to distension of the splenic capsule or space-occupying mechanisms within the abdominal cavity, or they may present as a palpable mass.[3,6] Symptoms secondary to pressure on surrounding organs, such as nausea, vomiting, flatulence, and diarrhea may gradually appear. Also, pressure in the cardio-respiratory system may cause pleuritic pain or dyspnea, and irritation of the left diaphragm may cause persistent cough. Occasionally, splenic cysts may present with complications, such as infection, rupture and hemorrhage.[4,5]

During the clinical examination, when a palpated mass is detected in the left upper quadrant of the abdomen, it is necessary to exclude any disease associated with splenomegaly, mononucleosis and fever of unknown origin, hemolytic anemia, chronic

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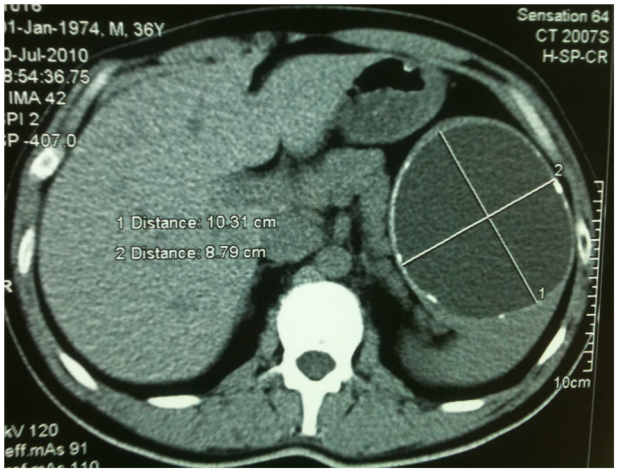

Fig. 1. A solitary, smooth outlined cyst, with signs of calcifications, occupying almost the whole splenic parenchyma, and particularly the splenic hilum (patient A).

leukemias, collagen vascular diseases, and liver diseases. Serological studies are useful in excluding most of the above-mentioned diagnoses.[7] Angiography is useful in differentiating a splenic cyst, which is usually avascular, from solid malignant tumors (lymphomas, sarcomas), which usually are characterized by neoplastic vasculature in a disorganized pattern.[8] Ultrasound examination is able to determine if the cysts are either anechoic or hypoechoic and if they are surrounded by a smooth thin wall.[8] In contrast, solid tumors are either isoechoic or hyperechoic in ultrasound imaging. Additionally, CT and magnetic resonance imaging (MRI) may provide most of the necessary information, regarding the morphology of the cyst, the composition of the cystic fluid, the location in the spleen, the position of the cyst and its relationship with the surrounding tissues.[2,8] Calcifications of both the primary and secondary cysts are frequently found, which are useful in diagnosing cysts from other causes of splenomegaly.

Definite diagnosis of the SP is set with the help of the histologic examination, where the lack of epithelial lining, the thickness of fibrotic wall and the detection of calcifications within the wall, point toward a non-parasitic, false cyst.[3] The cytologic examination of the content of the cyst will reveal the absence of malignant cells and parasites.

In this study, we present two unusual case reports of patients with an oversized splenic pseudocyst and we are introducing an update on current management for large SPs through literature.

2. Presentation of case

2.1. Patient A

Patient A was a 36 years old male with a history of abdominal injury in the past. He came to the emergency room with symptoms of abdominal pain and dyspepsia. The basic laboratory tests were non specific. The ultrasound examination showed a large, solitary, cystic mass of the spleen. The serological tests were negative for antiechinococcal antibodies. The patient was scheduled for CT control of the upper abdomen which revealed a solitary, smooth outlined splenic cyst, with signs of calcification. The cyst was occupying almost the entire splenic parenchyma, and particularly the splenic hilum, and it was 10.31 cm × 8.79 cm in size (Fig. 1). All data, so far, including the history of a past injury, supported the diagnosis of a SP.

Patient A underwent a laparotomy and total splenectomy. The histologic examination showed a splenic pseudocyst with a thick, fibrotic wall, and sparse calcifications within the cystic wall, but

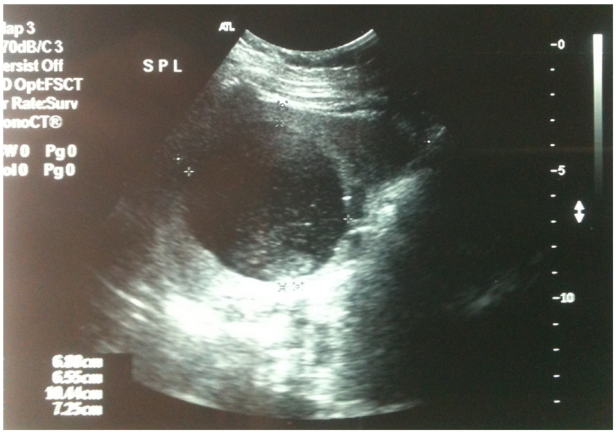

Fig. 2. A large solitary cyst of the spleen. Ultrasound examination of patient B.

lacking any epithelial elements. The liquid contained in the cyst was sent for a cytologic examination, which was negative for malignant cells or parasites. The patient underwent a full vaccination protocol, for avoidance of postoperative infections. Six months after the surgical procedure, no infections or other complications have been observed.

2.2. Patient B

Patient B was a 39 years old female with a history of ulcerative colitis. She came to the emergency room complaining for a painful mass in her left upper abdomen. The basic laboratory tests were in this case not specific as well. The ultrasound examination showed a solitary, cystic mass of the spleen of great size (Fig. 2). The serological tests were negative for antiechinococcal antibodies. The findings of the CT control revealed a solitary, smooth outlined cyst, with signs of calcification, occupying most of the splenic parenchyma (Fig. 3). The cyst was measured 7 cm × 7 cm. The possible diagnosis, so far, was a SP of unknown origin, since there was no obvious history of infarction, infection or injury in the past.

Patient B underwent a laparotomy with a total splenectomy, as well. The histologic examination described a splenic cyst with a thick, fibrotic wall, but without epithelial elements. Calcifications were also observed within the cystic wall, but in a lower density compared to patient A. The cytologic examination of the cystic liquid was negative for malignant cells or parasites. Full vaccination was performed according to protocol. One year after surgery, no

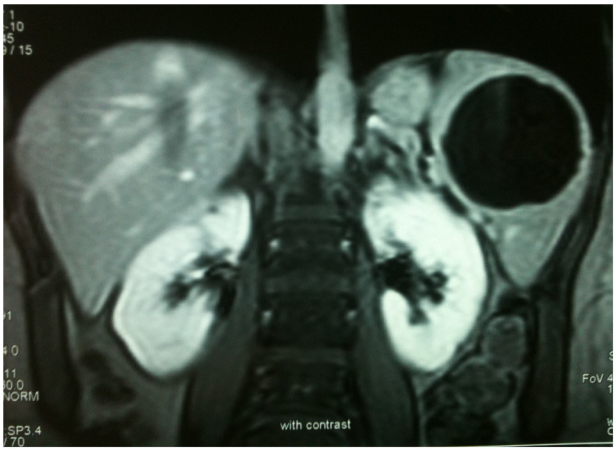

Fig. 3. Solitary, smooth outlined cyst, occupying most of the splenic parenchyma (patient B).

adverse events such as infection or other complications have been observed.

3. Discussion

As mentioned above, the hematological, biochemical, and serological investigations in both our patients with a solitary splenic pseudocyst were negative, leading to the definitive diagnosis of a non-parasitic pseudocyst of the spleen. Moreover, both patients presented with symptoms, although the majority of patients with splenic cysts are asymptomatic. In our cases, both ultrasound and CT examination set the diagnosis of a solitary unilocular splenic cyst that covered almost the entire splenic parenchyma. The cysts of both patients had smooth boundaries and a calcified lining, which covered most of the cystic wall. The medical history confirmed a previous abdominal injury only in case of patient A, whereas the history of patient B did not reveal any information leading to the cause of the pseudocyst development.

Concerning the management of splenic cysts, due to the increased risk of complications, splenic cysts with a diameter larger than 4–5 cm, as in patient A and B, should be managed surgically.[9,10] Additionally, more conservative options of splenic cyst management, such as percutaneous aspiration or infusion of sclerotic substances, do not result in long-term control.[11]

There are different types of surgical treatment according to the patient's age and the size, location and nature of the cyst. The classical approach to splenic cysts for years has been the open total splenectomy.[2,9,11] However, a new trend was introduced toward more conservative surgery after the 1970s, because of the appearance of overwhelming life-threatening septicaemia and increased prevelance of postoperative infections, especially in children who underwent splenectomy.[12,13] Indeed, the spleen plays an important role in hematopoiesis, immune function, and protection against infections and malignancies. Today, treatment options include partial splenectomy, total cystectomy, marsupialization, or cyst decapsulation (unroofing), accessed either by open laparotomy or laparoscopy.[12–15]

Concerning partial splenectomy, this method preserves more than 25% of splenic parenchyma, which is the minimal splenic tissue needed to preserve immunologic protection without increasing the risk of recurrence.[16,17] Furthermore, partial splenectomy can be performed safely with the laparoscopic approach.[18] This procedure is recommended, if the cyst is located in the poles of the spleen, or if the cyst cavity is deep, due to the higher risk of recurrence. Incision of the splenic capsule and hemostasis is performed with the ultrasonic or the monopolar scissors. A more conservative option could be a partial cystectomy (unroofing) of the cyst.[19] However, it has yet to be determined how much of the cyst wall should be resected, and whether unroofing should be partial or radical. It is supported that as much of the cyst wall as possible should be resected to prevent closure of the cyst. The marsupialization of the cyst is another conservative option recommended for superficial splenic cysts, and can be performed safely with the laparoscopic method.[20] This approach reduces the duration of the operation and carries no risk of recurrence.

In general, the laparoscopic management of splenic cysts offers the benefits of minimally invasive surgery: minimal postoperative pain, faster recovery, shorter hospital stay, and reduced morbidity and recovery.[18–20] However, any type of conservative procedure is difficult to perform, if (a) the cyst is oversized covering almost the entire splenic parenchyma, (b) is located in the splenic hilum, (c) is covered completely by the splenic parenchyma (intrasplenic cyst), or (d) if there are multiple cysts (polycystic cases). In our cases, there was an open total splenectomy performed, because, according to CT findings, a partial splenectomy was not indicated. The radiological examination of both patients revealed a splenic cyst of such great size (oversized cyst), that there was no possibility of preserving enough splenic parenchyma (at least 25%) in order to perform a partial splenectomy. Furthermore, the splenic cyst of patient A was occupying the splenic hilum and therefore, total splenectomy was the only surgical treatment option.

Regarding the postoperative course of our patients, no complications have been reported so far. Although partial splenectomy, either open or laparoscopic, has prevailed the last years, open total splenectomy ensures total cyst removal, avoidance of cyst-related complications and no cyst recurrence. Any type of spleen-sparing procedure (especially partial splenectomy) is not easy to perform in cases of surgical and anatomical difficulty, as mentioned above, because of recurrence and the risk of intractable bleeding from the spleen. Moreover, a well defined postoperative protocol of vaccination reduces the risk for severe sepsis and complications.

As a result, open total splenectomy should be proposed for symptomatic oversized splenic cysts and can be performed safely without any postoperative adverse events.

4. Conclusions

Symptomatic oversized pseudocysts of the spleen should be removed with open total splenectomy in order to remove the cyst with safety or when the size and location of the cyst do not allow the preservation of splenic parenchyma.

Conflict of interest

There was no conflict of interest.

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Ethical approval

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

Author contributions

George Galyfos: study concept and design, writing the paper. Zisis Touloumis: study concept and design, data interpretation. Konstantinos Palogos: data analysis and interpretation. Konstantinos Stergios: data collection and reference analysis. Maria Chalasti: data collection and analysis. Nikolaos Kavouras: data collection, writing the paper. Laurant Lavant: supervision, final approval of the paper.

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