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Giant Splenic Mesothelial Cyst Treated by Minimal Access Surgery—Case Report

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Abstract

A 16-year-old girl with an asymptomatic large splenic cyst. The disease was discovered by an accidental ultrasound of the abdomen due to a urinary infection. The patient was followed up conservatively for 3 years, and after the gradual enlargement of the cyst, laparoscopic surgery was indicated. The operation and postoperative follow-up for a period of 6 months is satisfactory.

Keywords: spleen, cyst, laparascopy

1. Introduction

Splenic cysts are a rare condition in pediatric age and are usually asymptomatic. Symptomatic patients usually present with abdominal pain or rarely abdominal lump. Small asymptomatic splenic cysts which are less than 5 centimeters can be preserved. In recent times, the treatment of primary splenic cysts in children that are larger than 5 cm is conservatively surgical, i.e. laparoscopic with preservation of the spleen.

2. Case Report

16-old female patient referred to our Clinic for Pediatric Surgery presented due to an accidentally ultrasound-verified large cyst of the spleen. The cyst was noticed 3 years before the operation when it was incidentally detected by US of the abdomen, during the evaluation for a urinary infection. The patient did not complain of abdominal pain, vomiting, elevated body temperature, nor she did not have any symptoms related to a large cyst in the abdomen. Abdominal ultrasound and MRI were performed, which showed the following: In the area of the upper half of the spleen, a larger parenchymal cyst can be observed, with dense contents, encapsulated, diameter approx. 85x72x90mm... (Figure 1, 2)

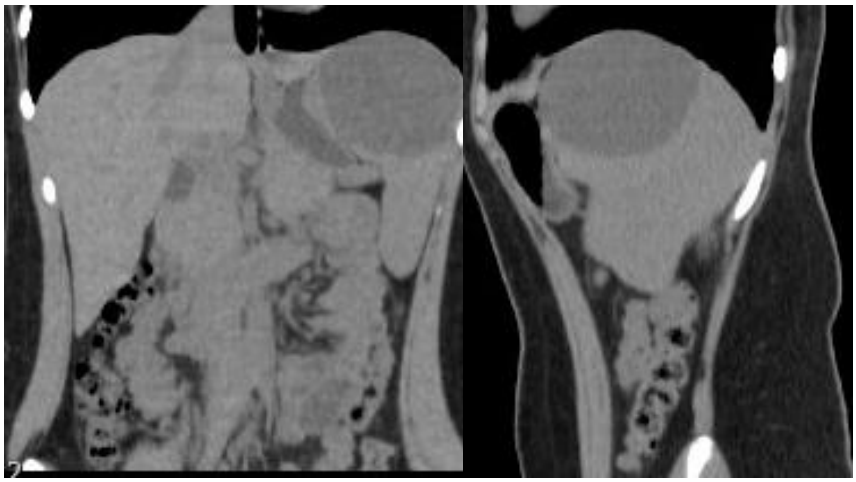

Figure 1 & 2. MRI In the area of the upper half of the spleen, a parenchymal cyst, with dense contents, encapsulated, diameter 85x72x90mm

The patient was conservatively monitored in the regional health institution. The ultrasound of the other abdominal organs was normal, and the X-ray of the lungs did not show the shadow of the cyst characteristics. Laboratory findings were also performed, inflammatory parameters were normal, tumor markers were negative. Due to the suspicion of the existence of an echinococcal cyst, we applied the diagnostic method ELISA test and due to the suspicion of the existence of an epithelial cyst, we performed the carbohydrate antigen 19-9 (CA 19-9). Both markers were negative. The patient denies a blunt abdominal injury in the earlier period of her life and also does not remember any severe febrile conditions that she experienced nor was she hospitalized due to a blunt abdominal injury or some unclear febrile conditions.

The patient was admitted to our clinic and after preoperative preparation, minimal access surgery was performed. The first optical port is introduced 120 mm subchondral, and a large cystic change in the area of the upper pole of the spleen is verified, which has not attached to the surrounding structures or to the diaphragm. Two more 5mm trocars are introduced, one in the medial line and one in the anterior axillary, both subchondrally. A cystic formation in the area of the upper pole of the spleen is verified. (Figure 3) The cystic formation is punctured and approximately 300 ml of yellow-greenish-hemorrhagic content is evacuated. (Figure 4)

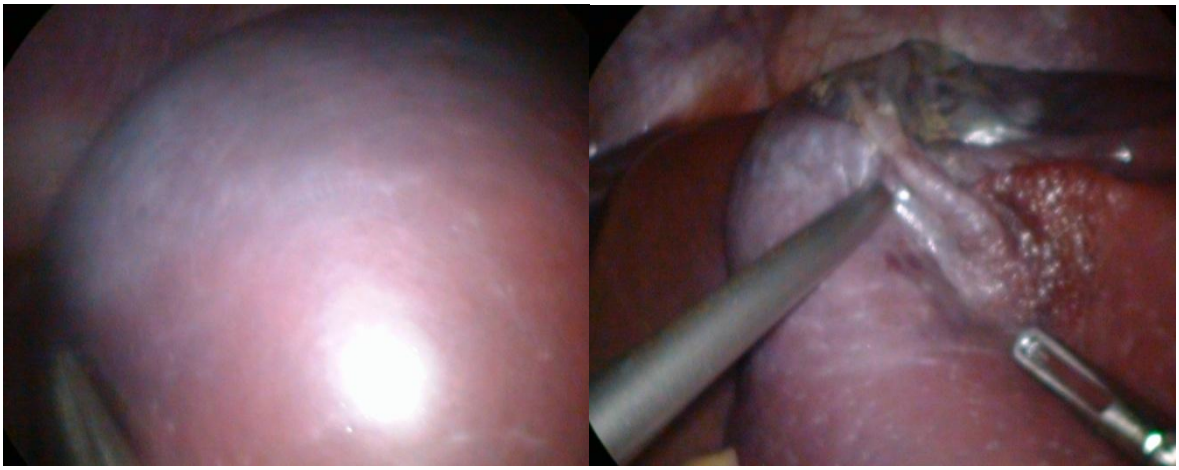

Figure 3 & 4. Cystic formation in the area of the upper pole of the spleen punctured

After the evacuation of the content, the boundary between the parenchyma of the lower pole and the thin cystic formation is clearly verified (Figure 5). Resection of the cystic wall, which protrudes above the spleen and has a thinned wall, is performed (Figure 6). Additional contents are evacuated. Careful hemostasis of the edge of the spleen is performed.

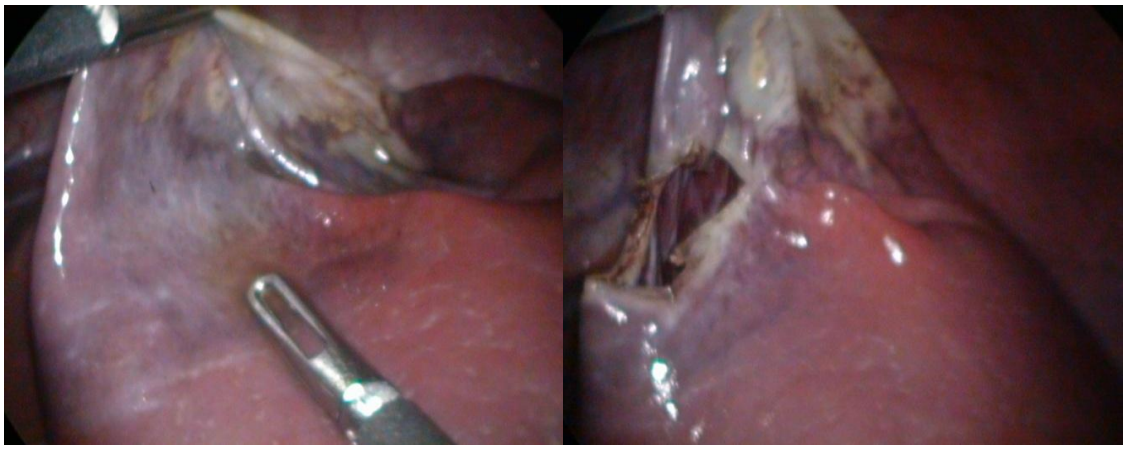

Figure 5 & 6. Resection of the cystic wall

The surface of the spleen, which formed the bottom of the cyst, did not appear to be covered with epithelium but with crypts and trabeculae corresponding to splenic tissue. (Figure 7, 8)

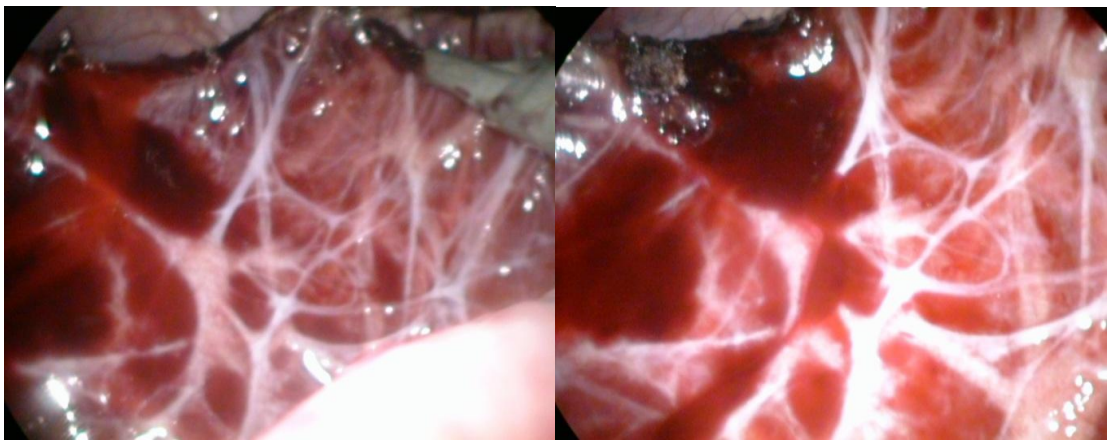

Figure 7 & 8. Surface of the spleen

After the removal of the cystic formation, a vulnerable surface remained in the area of the upper pole of the spleen. (Figure 9). Fibrospun tamponade is performed (Figure 10) and a parasplenic drain is placed (Figure 11).

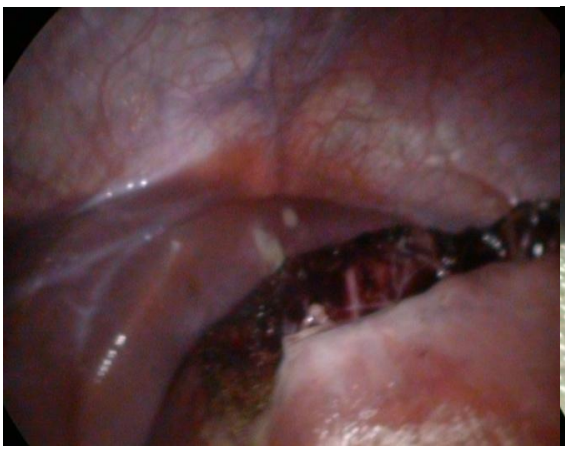

Figure 9. Vulnerable surface

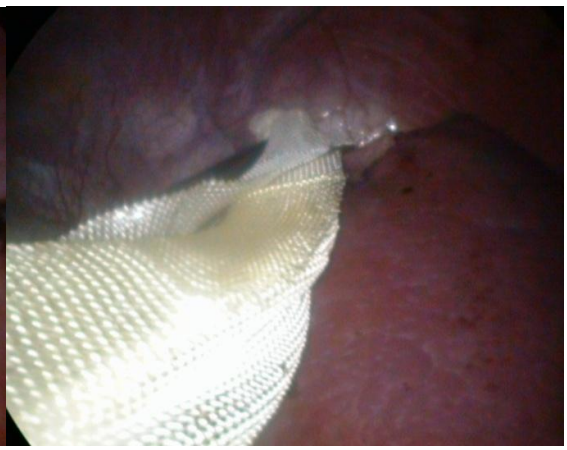

Figure 10. Fibrospun tamponade

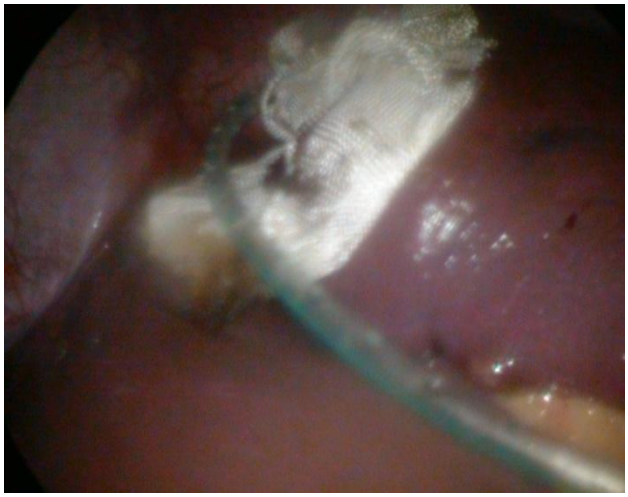

Figure 11. Parasplenic drain

The postoperative course is satisfactory. The patient is afebrile, peristalsis is established on time. Cephasollin a 1g x 3 is prescribed. Control laboratory findings are normal. The perisplenic drain is removed on the 3rd postoperative day. On the 4th postoperative day, laboratory findings are controlled: Le=7.4, CRP=48.6. The patient is discharged after that for home treatment. Regular ultrasound controls were performed and the last ultrasound control was performed 6 months after the operation and shows no signs of recurrence. After 6 months after the operation, the patient feels well and has no complaints. As part of the postoperative follow-up, we determined the values of CA 19-9 and CA 72-4 as a sign of possible recurrence of epithelial cysts of the spleen or the possible occurrence of mucinous adenocarcinoma of the pancreas, but they were negative.

The pathohistological findings of the wall of the splenic cyst indicate that it is most likely a mesothelial cyst: “The operative material consists of several fragments of irregularly shaped, grayish-brown, tougher tissue consistency.Microscopically, cyst wall patterns with remnants of splenic parenchyma. Cyst lined with single-row cuboidal epithelium, the morphological picture in the submitted material may correspond to a mesothelial cyst of the spleen”. (Figure 12, 13, 14).

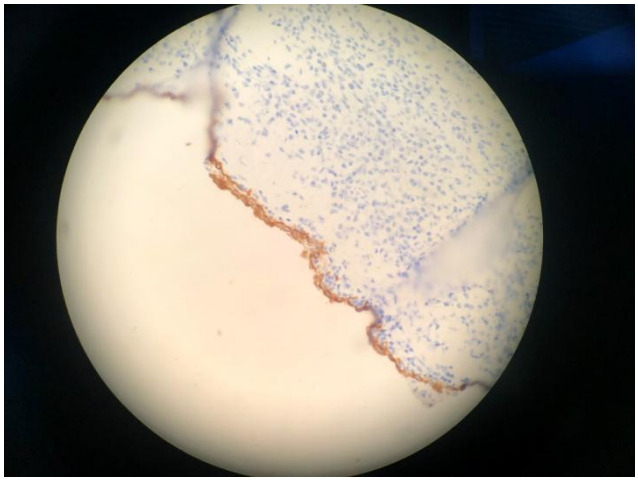

Figure 12. Immunophenotyping showed positivity for low molecular weight cytokeratin

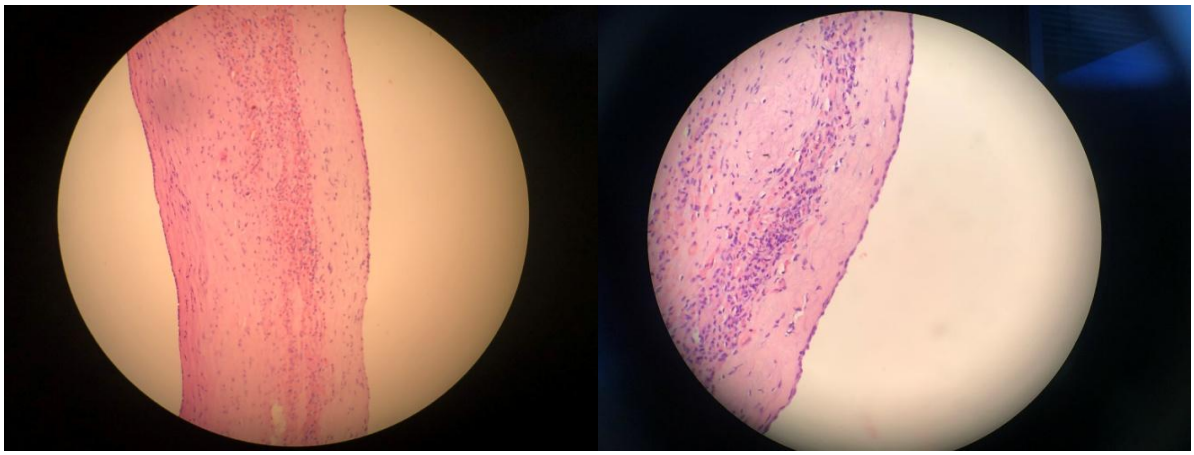

Figure 13 &14. Splenic tissue with cyst that was lined by flattened cuboidal cells (hematoxylin & eosin staining)

3. Discussion

Splenic cysts are rare with an incidence of 0.07%. in a review of over 42,000 autopsies (Morgenstern L, 2002). Andral, in 1829, first described a case of splenic dermoid cyst at an autopsy (Andral G, 1829). In 1974, Robbins et al1 reported only 32 cases of splenic cysts in 42 327 splenic autopsies during a period of 24 years (Robbins FG, Yellin AE, Lingua RW & et al., 1978). These splenic cysts were incidental postmortem findings.

The first classification of non-parasitic splenic cysts was published by Fowler (Fowler RH, 1953). He divided all cysts depending on the presence of epithelial lining into primary or true cysts and pseudocysts. Splenic cysts are currently categorized as parasitic (usually caused by Echinococcus granulosus and found in endemic areas) or nonparasitic.The classification is by Mirilas P et al. (Mirilas P, Mentessidou A & Skandalakis JE, 2007), who classified splenic cysts into primary splenic cysts (includes congenital and neoplastic cysts) and secondary cysts (consisting of traumatic and necrotic cysts. Mirilas are also like Fowler true cysts are also called congenital cysts and are defined by the presence of an inner endothelial lining and may be mesothelial, transitional, or squamous (Kawashima A & Fishman E., 1994). They are developmental in origin and the lining is formed secondary to an infolding of peritoneal mesothelium or collection of mesothelial cells trapped within splenic sulci (Freeman JL, Jafri SZ, Roberts JL, Mezwa DG & Shirkhoda A, 1993; Ough YD, Nash HR & Wood DA, 1981).

Although the exact mechanisms of its etiology, pathogenesis, and development are still unknown, proposed mechanisms include invagination of the mesothelial capsular surface with subsequent cyst formation, embryonic inclusion or metaplasia of epithelial cells of adjacent structures, as well as a mechanism involving peritoneal endothelial cells (Morgenstern L, 2002; Andral G., 1829; Robbins FG, Yellin AE, Lingua RW & et al., 1978). In pathological examinations the mesothelium is usually unilocular and varies in size. The surface is whitish or grayish-white, smooth, and shiny, and the inner portion exhibits coarse trabeculations due to subepithelial fibrosis. Palmieri et al. (Palmieri I, Natale E, Crafa F, Cavallaro A & Mingazzini PL, 2005) have established the pathological diagnosis of congenital cysts by immunohistochemistry using cytokeratin, CEA, CA19-9, and Calretinin. Secondary cysts tend to be the result of a trauma due to a failure in the organization of subcapsular or parenchymal hematomas; more rarely, they are the result of necrosis or abscesse. Primary cysts are less common than secondary cysts and account for approximately 30-40% of all splenic cysts. Congenital non-parasitic cysts account for approximately 25% of primary cyst cases, and are more common in children than in adults.

Most cysts are asymptomatic as it was in the case report of our 16-year-old girl who accidentally discovered a spleen cyst by doing an abdominal ultrasound after urinary infections.The usual clinical presentation of symptomatic splenic cysts includes left upper abdominal pain or discomfort often accompanied by feeling of fullness. Infected cysts can present with fever, vomiting or a raised leukocyte count. Splenomegaly is usually present when the cyst is more than 6 cm in size (Vijayaraghavan R, Chandrashekar R, Aithal S, Rashmi MV & Belagavi CS., 2010). Splenic cysts larger than 5 cm are more prone to complications like haemorrhage, rupture or infection and surgical treatment is therefore recommended (Zvizdić Z & Karavdić K., 2013). Control of the cyst size, prevention of complications, and avoidance of recurrence must be the basic principles of management of splenic cysts. (Hansen MB & Moller AC., 2004)

Different types of operative management of nonparasitic splenic cysts are available,including percutaneous drainage, complete splenectomy,partial splenectomy, marsupialization of the cyst, and partial cystectomy (fenestration, unroofing, deroofing). Factors like patient age and the size, location, and nature of the cyst influence the selection of the applied surgical procedure. (Gianoma D, Wildisenb A, Hotza T & et al., 2003)

The first attempt to excise a splenic cyst was performed by the French surgeon Jules Pe'an in 1867, but the attempt failed because of excessive bleeding, and a total splenectomy was performed. In cases with surgical indication and due to the growing

popularity of spleen preservation to preserve immune function, many alternative treatment options have been suggested. These include aspiration, internal and external marsupialization, partial splenectomy, partial cystectomy (decapsulation), and partial laparoscopic cystectomy Its advantages include a simpler and faster procedure with less blood loss. Disadvantages include the possibility of recurrence of the cyst, since a portion of the cyst lining is left intact. Cyst aspiration as definitive treatment has been described, but it has not been successful. Agents such as tetracycline or alcohol are injected into the cysts to destroy the lining, but cases of recurrence have occurred.This procedure is intended as only a temporary measure.Marsupialization is defined as the creation of an opening in the wall of the cyst in order to drain it. This opening may be internal (within the peritoneal cavity) or external (through an intentional cyst-cutaneous fistula). This treatment leads to cyst recurrence and is not recommended. Cystectomy with partial splenectomy consists of cyst resection with a contiguous part of the spleen parenchyma and was first reported by Morgenstern and Shapiro in 1980. Removing the cyst wall and its interior in their entirety is the only definitive treatment that can ensure that no remaining cyst tissue remains, leading to the absence of symptoms or recurrences, Morgenstern and Shapiro first performed partial splenectomy for splenic cyst (Morgenstern L & Shapiro SJ, 1980). Splenic preservation techniques include decapsulation, deroofing, marsupialisation, cyst excision and partial splenectomy with or without placement of an omental pedicle. Any or all of these procedures can be performed either by laparoscopic or open methods (Hansen MB & Moller AC, 2004; Williams RJ & Glazer G, 1993; Mackenzie RK, Youngson GG & Mahomed AA., 2004; Tagaya N, Oda N, Furihata M, Nemoto T, Suzuki N & et al., 2002). Technical details during splenic conservation includes ligation of polar vessels, splenic division, buttressing sutures and omental pedicle (Schier F, Waag KL & Ure B, 2007). Decapsulation involves a near total excision of the cyst and leaving a portion of the cyst wall contiguous with splenic parenchyma. Advantages include less blood loss and speed of operation. Recurrence appears to be low (13).

Salky first introduced laparoscopic fenestration of a nonparasitic nonparasitic splenic cyst in 1985 Salky B, Zimmerman M, Bauer J, Gelernt I & Kreel I., 1985) In laparascopic approach, only a segment of the cystic wall

is resected, creating an opening for communication between the peritoneal and cyst cavities. Coagulation of the remaining part of the cystic wall that was not removed due to possible bleeding is recommended. Coagulation should prevent possible recurrence. In our case, we did not perform coagulation of the surface of the remaining inner wall covering the spleen because it appeared to us to be a secondary cyst, which had no epithelium. (Posta CG., 1994) However, it was only after the operation that we received the pathohistologist's findings that that surface was covered with cubical epithelium and that it was most likely a splenic mesothelial cyst The main disadvantage of this procedure is the remaining risk of recurrence.Even if previous reports support the attachment of the omentum over the parenchyma defect as a way of reducing the risk of recurrence, we decided on Fibrospun tamponade of the splenic space. It has not yet been determined how much of the cyst wall should be resected and whether deroofing should be partial or radical. It is believed, however, that removing a large amount of the cyst wall will prevent cyst reclosure (recurrence). (Taragarona EM, Martinez J, Ramos C & Becerra JA., 1995; Cala Z, Cvitanovic B, Perko Z, Velnic D & Rasic Z., 1996; Feliciotti F, Sottili M, Guerrieri M, Paganini AM & Lezoche E., 1996) With laparoscopic and open fenestration, the risk of recurrence is the same. (Bove T, Delvaux G, Van Eijkelenburg P, De Backer A & Willems G., 1996) The laparoscopic technique enables the surgeon to determine the precise size and shape of the splenic cyst and to successfully and thoroughly remove the cystic wall, as we did with our patient.

4. Conclusions

-Superficially located splenic cysts in pediatric ages should be treated by laparoscopic marsupialization or fenestration.

-Laparoscopic spleen-preserving surgery must be the first goal of management of nonparasitic splenic cysts, because it provides all the benefits of minimally invasive surgery and offers adequate safety and acceptable results.

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