Case Report

Epidermoid Splenic Cyst in Children Treated by Laparoscopic Partial Splenectomy

Salihou Aminou Sadjo, Christelle Destinval, Sophie Amenan Kan Kouassi-Dria, Julie Lienard, Aline Ranke, Olivier Larmure, Nicolas Berte, Vladimir Gomola, Jean-Louis Lemelle

Department of Pediatric Surgery, Nancy Regional University Hospital, Nancy, Vandoeuvre-les-Nancy, France

Abstract

Epidermoid splenic cyst is a rare benign tumour in children, accounting for 10% of all cystic lesions of the spleen and is the most common diagnosis of non-parasitic splenic cysts. Its discovery could be incidental or happen after an imaging workup for a mass or abdominal pain. Since total splenectomy exposes a patient to the occurrence of overwhelming post-splenectomy infection, laparoscopic partial splenectomy has been increasingly popular over the past two decades because it preserves the immune function of the spleen. This technique should only be performed by a well-trained team because of the high risk of bleeding. The authors report the cases of three patients who underwent laparoscopic partial splenectomy: a 7-year-old girl with no relevant history with a 4.5-cm epidermoid cyst involving the lower pole of the spleen, a 13-year-old boy, type 1 diabetic on insulin therapy, with a 7-cm epidermoid cyst of the upper pole of the spleen and a 14-year-old girl, with no previous history, with a 6-cm upper pole epidermoid splenic cyst. Operating times were 2 h 30 min, 3 h and 4 h 30 min, respectively. The intraoperative blood loss was 100 mL, 350 mL and 300 mL, respectively. The length of hospital stay was 6 days for each patient. No blood transfusion was performed perioperatively. With a mean follow-up duration of 21 months (32 months, 21 months and 10 months, respectively), no complication or recurrence occurred.

Keywords: Children, epidermoid cysts, laparoscopy, partial splenectomy

Introduction

First described in 1829 by Andral epidermoid splenic cyst is a rare clinical entity in current paediatric surgical practice.[1] Robbins *et al*. in the United States reported 23 cases out of 42,327 autopsies. It is most often an incidental finding (incidence of 0.07%), but it can also be discovered during the assessment for chronic abdominal pain located on left hypochondrium.[2] Surgical approach of epidermoid splenic cyst in children first described by Andra was total splenectomy.[1] Splenic preservation has progressively become the goal standard, first introduced by Morgenstern to preserve immune function.[3] Laparoscopic approach for partial splenectomy in children has been increasingly popular over the past two decades. This technique should only be carried out within a well-trained team because the risk of bleeding is high. Total splenectomy may lead to an overwhelming post-splenectomy infection (OPSI). We report three cases of epidermoid splenic cyst treated in our surgical department by partial laparoscopic splenectomy and reviewed cases from the literature.

Case Reports

Case 1

A 7-year-old girl without medical history was brought in the emergency department for recurrent left abdominal pain running for 1 year. No fever, vomiting, or transit disorders was disclaimed. Physical examination found isolated tenderness of the left hypochondrium. Abdominal ultrasound (US) showed a splenic cyst located at the lower pole of the spleen, with a 4.5-cm uncalcified wall. Abdominal magnetic resonance

Received: 07-07-2023 **Revised:** 15-08-2023 **Accepted:** 06-11-2023 **Available Online:** 13-09-2024

Access this article online

Quick Response Code:

Website: https://journals.lww.com/AJPS

DOI: 10.4103/ajps.ajps_84_23

Address for correspondence: Dr. Salihou Aminou Sadjo, Department of Pediatric Surgery, Pediatric Surgery Service, Nancy Regional University Hospital, 1 Rue du Morvan, Vandoeuvre-les-Nancy 54500, France.
E-mail: sas.amine2@yahoo.fr

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How to cite this article: Sadjo SA, Destinval C, Kouassi-Dria SA, Lienard J, Ranke A, Larmure O, *et al*. Epidermoid splenic cyst in children treated by laparoscopic partial splenectomy. Afr J Paediatr Surg 2024;21:278-82.

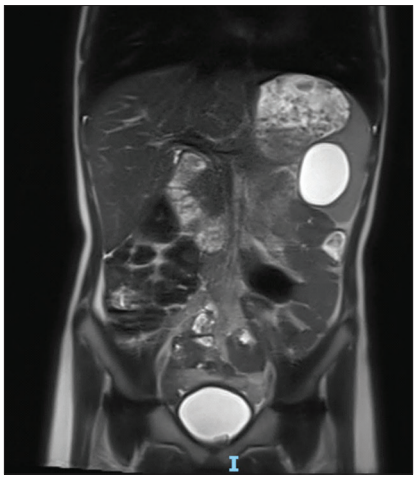


Figure 1: coronal magnetic resonance imaging of the child abdomen showing well-defined cystic mass with homogeneous fluid contents (Case 1)

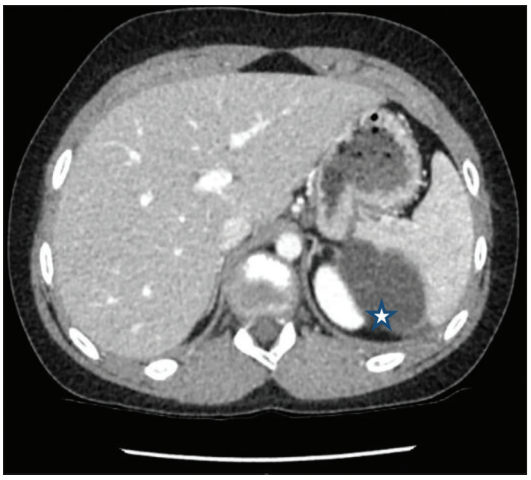


Figure 2: Scan showing a hypodense cystic image within the spleen (white star)

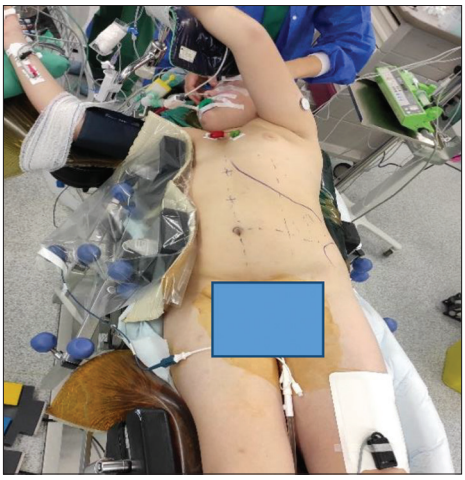


Figure 3: Patient installed in a 3/4 right lateral decubitus position with a log under the scapula and the left buttock

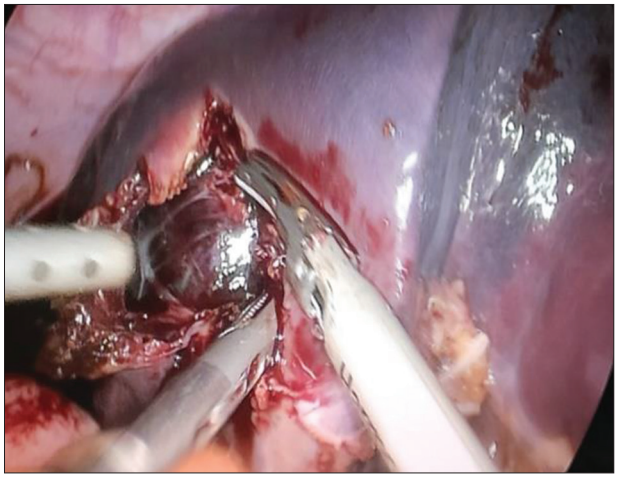


Figure 4: Vascular control with Hem-o-lok

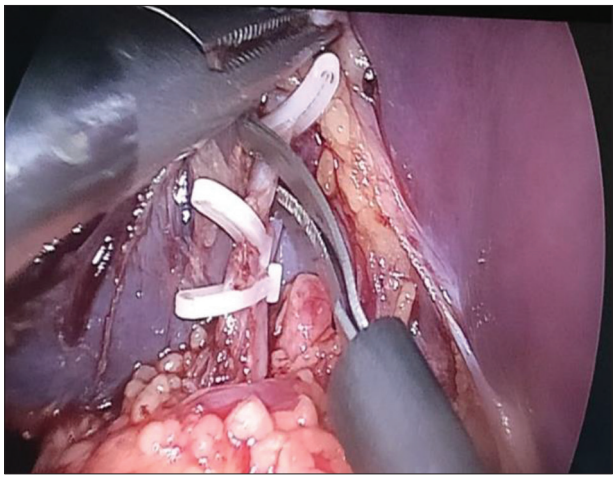


Figure 5: Opening and aspiration of the cyst



Figure 6: Application of TACHOSIL® gauze on the section area

imaging (MRI) highlighted an anechogenic mass related to the splenic hilum without calcification [Figure 1]. No sign of compression was described. Routine biology was normal and so was hydatid serology. As recommended, pre-operative immunisation was practiced (against pneumococcus, meningococcus and *Haemophilus influenzae*). Inferior partial splenectomy was performed by laparoscopy. Blood loss was

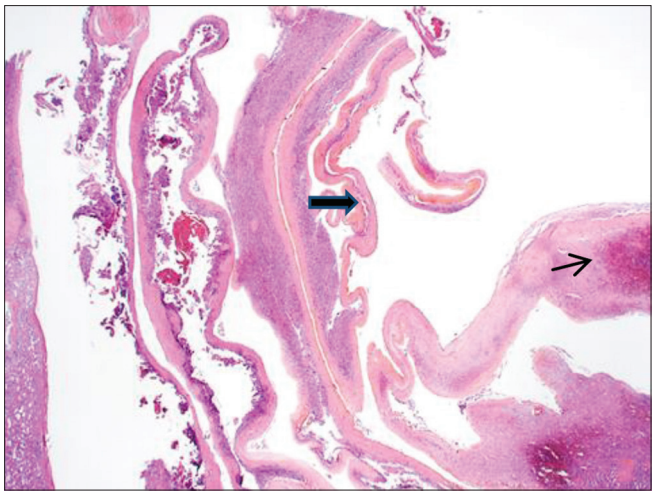


Figure 7: HES with low magnification. Splenic parenchyma (→) shifted peripherally by a cystic lesion (➔)

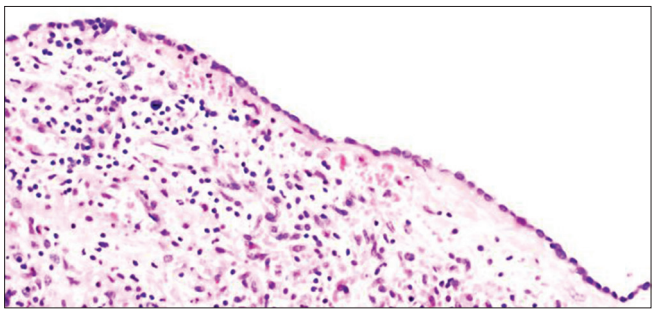


Figure 8: HES with high magnification. The lining of the cyst is a flattened cuboidal epithelium of mesothelial type

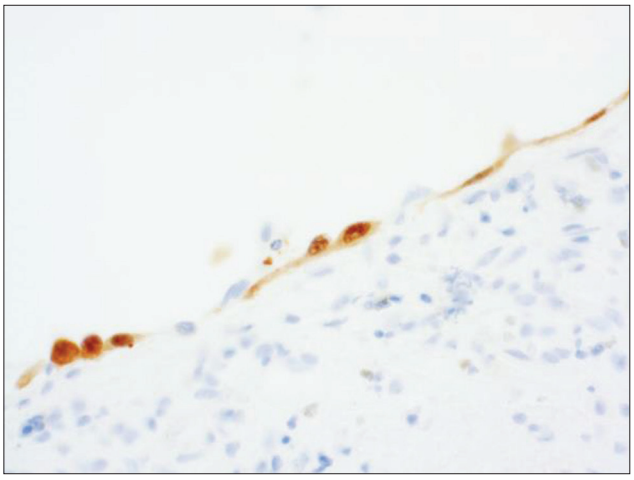


Figure 9: Immunohistochemistry. The anti-calretinin antibody highlights the epithelium confirming the mesothelial nature of the cyst

about 100 mL. Blood transfusion has not been required and no perioperative complication was described. Operative time was 2 h 30 min. Post-operative course was free of complication and hospital length of stay was 6 days. A 3-month complete sport discharge was prescribed. Antibiotic prophylaxis was introduced by roxithromycin for 1 year because of ampicillin allergy. Histopathology confirmed epidermoid origin of the cyst. One year later, abdominal US showed good vascularisation of the rest of the spleen and allowed discontinuation of antibiotics. After a 32-month follow-up duration, no complication or recurrence was recorded.

Case 2

A 13-year-old male, diabetic (type 1) with an insulin pump for 6 years, consulted for isolated abdominal pain in the left hypochondrium. Abdominal US revealed a 2-cm cyst in the upper pole of the spleen. Surgical resection was not proposed at initial management because of the cyst length (<5 cm). Analgesics helped relieving the pain for a time, but recurrence and intensification of the pain motivated a second consultation 4 years later. Abdominal computed tomography (CT) scan noted an increase of the cyst diameter to 7 cm, without enhancement after injection in favour of an epidermoid cyst [Figure 2]. Biological examinations were normal, including hydatid or other parasitic serology. Pre-operative immunisation was administered. Laparoscopic partial splenectomy was performed with an estimated blood loss of 350 mL. No blood transfusion was necessary. Operative time was 3 h. On post-operative day 2 (POD 2), fever (38, 5°C) and right cervical swelling revealed parotitis which was confirmed on cervical US. Amoxicillin–clavulanic acid was administered for 10 days, with good efficiency and tolerance. Discharge home was possible on POD 6 with a 3-month complete sport discharge. Pathological findings were in favour of epidermoid cyst. Antibioprophylaxis by amoxicillin was taken for 1 year. A 21-month follow-up duration did not show any recurrence or complication and is still ongoing.

Case 3

A 14-year-old girl, with no relevant medical history, was hospitalised for a non-febrile gastroenteritis. An abdominal US was performed for persistent abdominal pain localised in the left hypochondrium and showed an 8-cm splenic cyst protruding into the hilum. Imaging assessment was completed by an abdominal CT scan that confirmed the diagnosis of splenic cyst located at the superior pole of the spleen. Biological investigations excluded parasitic or tumour origin. Pre-operative immunisation against pneumococcus, meningococcus and *H. influenzae* was proceeded. We performed a partial splenectomy of the upper spleen by laparoscopy. Operative time was 4 h 30 min and a 300 mL blood loss was estimated. The post-operative period was out of complication and discharge was possible on POD 6 with a 3-month complete sport discharge. Histopathology confirmed the diagnosis of epidermoid cyst. Antibioprophylaxis by amoxicillin was administered for 1 year. A 10-month follow-up duration showed no complication or recurrence.

Surgical technique

Anaesthetic considerations were classic endotracheal intubation, general anaesthesia associated to locoregional anaesthesia (paravertebral bloc), one arterial catheter and two venous access (because of the high risk of bleeding).

Installation was right semilateral decubitus with log under the scapula and the left buttock [Figure 3]. Open laparoscopy was made by a short periumbilical incision for the optical port. CO_2 pneumoperitoneum was created with insufflation pressure ranging from 8 mmHg (Case 1) to 10 mmHg (Cases 2 and 3). Two 5-mm diameter operator ports were placed (one in the left hypochondrium and the other in the epigastric region). A 5-mm diameter third one was placed below the xyphoid process for liver retraction. In Case 3, a 10-mm port was added directly above the spleen. Access to the lesser omental and to splenic hilum was realised by a left-sided approach based on gastrocolic, splenocolic and gastrosplenic ligaments dissection. Mobilisation of the spleen was realised by sectioning short gastric vessels using ultrasonically activated scalpel (ULTRACISION®) or ligature vessel sealing system (LVSS) depending on surgeon preference. The upper and lower branches of the splenic pedicle were dissected. Arterial and venous control was secured with plastic Hem-o-lok® clips before section [Figure 4]. At this stage, an ischaemic demarcation line was observed and became the dissection guideline. Cyst dome opening helped to facilitate dissection in Case 2 [Figure 5]. Transection of splenic parenchyma was performed using ULTRACISION® (Case 2) or LVSS (Cases 1 and 3), at about 1 cm from the dissection guideline [Figure 6]. Haemostasis was achieved by bipolar coagulation and collagen sponge coated with human coagulation factors fibrinogen and thrombin (TACHOSIL®) in all cases. Resected part of the spleen was exteriorised in an Endobag™ retrieval system throughout the umbilical port incision. No drainage was left.

Discussion

Splenic epidermoid cyst is a rare clinical entity in children. It preferentially affects adolescents and young adults, as much boys as girls.[4,5] Ages of cases in our short series are in line with cases in the literature.[5,6] Most often, a splenic epidermoid cyst is asymptomatic and diagnosed incidentally during an abdominal US. For Sinha and Agrawal, abdominal pain located on the left hypochondrium was the main manifestation (46% of cases) as found in our patients (100%).[7] Due to the location of pain in the left hypochondrium, possible differential diagnoses are gastritis, pancreatitis, colonic diverticulitis and epiploic appendagitis. It can also be raised by abdominal mass like in Garg *et al*. series.[5] Forms complicated by intraperitoneal rupture, haemorrhage or infection are exceptional, representing 1%–3% depending on the series.[7,8] No case of malignant degeneration has been reported in the literature to our knowledge. Imaging evaluation is crucial for the diagnosis. US is the first-line examination showing an anechogenic mass without calcification. It is most of the time completed by abdominal CT scan or MRI regarding cystic partitioning, calcification of the wall and relations with neighbouring organs. Usual management includes parasite serology.[7] Tumour markers concerned by splenic mass are carcinoembryonic antigens and carbohydrate antigen 19-9 that are slightly secreted by epidermoid cyst. They have been evaluated in all our three cases even if some authors recommended it when splenic lymphangioma is suspected.[5] It can also be used for post-operative monitoring.[3,9] Management of splenic epidermoid cyst is dependent on size, localisation and symptoms. Abstention is recommended by most authors in case of small asymptomatic cyst (<5 cm).[7,10] In other cases, surgical approach is required and can be cyst resection, cyst fenestration, partial splenectomy or total splenectomy. Cyst resection or fenestration has been recommended for many years, but progressively abandoned due to high rate of recurrences ranging 49% of cases.[7] However, it can be required in case of central localisation of the cyst, in splenic hilum that would contra indicates partial splenectomy. Except for these rare cases, partial splenectomy, described first in 1980 by Morgenstern, has become the gold standard.[3] Total splenectomy has been occasionally described but is more and more abandoned. Progress made on partial splenectomies for other indications (sickle cell anaemia, thalassaemia and autoimmune hepatitis with splenic sequestration) are applied for epidermoid cysts. Total spleen resection led to immunity dysfunction and imposes preventive antibiotics on the long term. Mortality of total splenectomy due to OPSI stays high ranging 50%–70%.[11] Total splenectomy is a radical procedure that should be considered for malignant tumours, for giant forms invading majority of the splenic parenchyma,[10] or for hilum localisation without surgical possibilities of partial resection. The advantages of partial splenectomy are preservation of immune functions authorising short antibioprophylaxis.[3] After 1 year, splenic function can be evaluated by *Howell Jolly's bodies* dosage, Doppler US of the remaining spleen or spleen scintigraphy. Only splenic Doppler US was performed in our patients postoperatively, scintigraphy being an irradiating examination and the search for *Howell Jolly's bodies* not being specific to functional hyposplenism. Another advantage of partial splenectomy for epidermoid cyst compared to cyst opening or resection is a lower rate of recurrence representing < 2% (1 of 52 cases) according to Sinha and Agrawal.[7] None of our cases developed recurrence, respectively with 32 (Case 1), 21 (Case 2) and 10-months follow-up duration (Case 3). Laparoscopic approach for this procedure has spread across the world over the past two decades. Haemorrhagic risk is the main intraoperative complication of this technique, justifying preventing patient of the risk of total splenectomy and proceeding in pre-operative immunisation.[7] Perioperative blood loss was acceptable in our results, as reported by Ramia *et al*. who described 130 cm^3.[12] No blood transfusion was required in our series. The length of hospital stay of 6 days in our patients, compared to 3 days reported by Jain *et al*.[13] is explained by the progressive mobilisation of our patients on one hand and on the other hand by the appearance of post-operative parotitis in the diabetic patient. Operative time in our series was acceptable from 2 h 30 min (Case 1) to 4 h 30 min (Case 3). Jain *et al*. described shorter time in his case report (1½ h).[13] It depends on technical difficulties,

localisation of the cyst, size of the cyst and surgeon experience. No complication, no conversion and no total splenectomy were recorded in our series. Definitive diagnosis can only be established on histopathological data by the presence of a cuboidal epithelial lining [Figures 7 and 8]. The anti-calretinin antibody highlights the epithelium, confirming the mesothelial nature of the cyst [Figure 9].

Conclusion

Laparoscopic partial splenectomy is the standard treatment for epidermoid splenic cyst in children. It can be safely performed when the surgical team is accustomed to laparoscopy. It has the advantage of preserving the immune function of the spleen and improving the post-operative comfort of the patient. Since total splenectomy exposes to the occurrence of overwhelming post-splenectomy infection (OPSI).

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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