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Management of nonparasitic splenic cysts in children: A French multicenter review of 100 cases☆,☆☆

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ABSTRACT

Background: The management of nonparasitic splenic cysts (NPSC) remains controversial. Surgical resection is indicated for symptomatic or complicated forms, but no guidelines are available for asymptomatic NPSC. The aims of this study were to evaluate the management of NPSC in French hospitals and to analyze the results of management.
Methods: We conducted a retrospective multicenter study from January 2004 to December 2014 in 16 university hospitals in France. Patients with a follow-up less than 6 months were excluded. Data were extracted from the medical reports.
Results: One hundred patients were included. Median follow-up was 12.8 months. No complications were observed for NPSC smaller than 5 cm. The size of NPSC increased significantly between the ages of 10 and 12 years. Fifteen patients were under observation; 58.3% of cysts decreased in size and 41.7% remained stable. Among the 85 operated patients, no recurrence occurred in the splenectomy group, while 11 recurrences were observed in the cystectomy group (57.9%), 3 of which required redo surgery.
Conclusions: Observation is a safe treatment option for asymptomatic NPSC smaller than 5 cm. Surgery is indicated for symptomatic patients, and can be proposed for asymptomatic NPSC larger than 5 cm. Laparoscopic partial splenectomy is the technique of choice. Follow-up must be continued until the end of puberty.
Levels of evidence rating: Level III.

Nonparasitic splenic cysts (NPSCs) are rare, as less than 1000 cases have been reported in the literature. However, all pediatric surgeons may encounter this diagnosis. No guidelines have been published concerning the management of these lesions in the pediatric population, and published series are often based on small numbers of patients.

NPSCs often present as asymptomatic lesions, discovered on systematic imaging or prenatal ultrasonography (US). However, NPSC can present in the form of complications, such as intracystic hemorrhage, cyst infection, or cyst rupture, requiring surgery. The management of nonparasitic splenic cysts remains controversial, especially for asymptomatic forms. As NPSC with a typical US presentation are benign lesions, treatment decisions are not impacted by the risk of malignancy.

The goal of management is to avoid complications. Since the publication by Péan and Magdelain [1], total splenectomy has been considered to be the gold standard for the treatment of splenic cysts. However, total

Abbreviations: US, ultrasound; CT, computed tomography; NPSC, nonparasitic splenic cyst.

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splenectomy is associated with a risk of serious infectious complications such as streptococcal septicemia. Physicians are now encouraged to avoid this procedure whenever possible [2]. A more conservative attitude has been proposed with the development of partial splenectomy [3,4], cystectomy or unroofing. All these techniques can be performed by laparoscopy [5]. The aims of the present study were to determine the surgical indications for asymptomatic NPSC, and to evaluate the results of the various surgical techniques, in terms of complications and recurrence.

We report the results of a French retrospective multicenter study based on a series of 100 patients.

1. Materials and methods

In this retrospective multicenter study, a questionnaire was sent to all French pediatric surgery departments to evaluate their management of NPSC and their results. Sixteen university hospitals in France and one university hospital in Brussels participated in this study. We retrospectively reviewed the patients' charts and collected data from consultations, imaging, and operative reports.

Patients with a diagnosis of splenic cyst between January 2004 and December 2014 with a follow-up of at least six months after diagnosis were included. Patients with parasitic lesions were excluded. The diagnosis of hydatid disease was suspected in cases of stay in endemic area or hypereosinophilia. Confirmation was obtained by echinococcosis serology, or postsurgical histopathology. Operated patients were identified by the national coding procedure. Nonoperated patients were identified by the surgeons. Clinical and imaging data were extracted from the patient's charts. The circumstances of discovery and age at diagnosis, cyst dimensions and variation in size in the case of nonsurgical treatment were recorded. Patients undergoing surgery confined to the cyst (cystectomy or unroofing) were classified in the “cystectomy” group. Patients undergoing partial or total splenectomy, at first-line or redo surgery, were classified in the “splenectomy” group. Nonoperated patients were classified in the “observational management” group. Perioperative complications and cyst recurrences were collected to evaluate surgical results.

Laparoscopic partial splenectomies were performed by an anterior approach. The patient was placed in right semilateral decubitus position. Surgeon and assistant were in front of the patient. The optic trocar was inserted in the umbilicus. One trocar was placed in the left lower quadrant, and one or two additional trocars were spread around the costal margin. This anterior approach provided a clear exposure of the hilum and splenic vessels, and allowed their selective division. After ligation of the concerned vessels, the spleen was divided along the edge of the ischemic part, using either a stapler or harmonic scalpel. In cases of laparoscopic total splenectomy, a lateral approach was performed. The patient was placed in the same position. Two additional ports were inserted in the epigastrium and in the left flank. Splenic dissection began at the lower pole of the spleen. The posterior splenophrenic attachment was divided, allowing the spleen to roll away from the lateral abdominal wall and thus exposing the splenic hilum and the tail of the pancreas. In this lateral approach, the direct access to the splenic vessels is well-adapted for total splenectomy.

Statistical analysis was performed with Graphpad Prism Software (La Jolla). Results are expressed as the mean and standard deviation (M ± SD). A Kolmogorov–Smirnov test was used to check the normal distribution of quantitative values. Groups were compared by chi-square test, and Fisher's exact test for smaller sample sizes. Continuous variables were compared using a Student *t*-test. A $p < 0.05$ value was considered statistically significant.

2. Results

One hundred five patients managed for nonparasitic splenic cyst in 16 university hospitals were studied. One experienced surgeon per center was in charge of the management and surgery of these patients. Five patients were excluded because of missing data. Short-term results were analyzed for 100 patients.

Except for cases of prenatal diagnosis, the mean age at diagnosis was 11.6 years (±0.3) (range: 3.8–17.5) with a balanced sex, with 48% of female patients. Median follow-up was 12.8 months (range: 6.1–101.7 months).

The circumstances of discovery were: incidental finding (43%), abdominal pain (32%), abdominal mass (10%), complicated form (7%), prenatal diagnosis (6%), and various abdominal symptoms (2%). Complicated forms consisted of intracystic hemorrhage in 4 patients and cyst rupture in 3 patients. Mean cyst diameter at diagnosis was 88.7 mm (±5.3) (range 7–265 mm). Asymptomatic cysts had a mean diameter of 67.9 mm (±6.7) and were significantly smaller than symptomatic cysts: 108.6 mm (±7.2) ($p < 0.01$). Complicated cysts had a mean diameter of 97.6 mm (±13.9) ($p < 0.01$). The proportion of symptomatic and complicated cysts increased with increasing cyst diameter. More than one-third of cysts larger than 5 cm were symptomatic and complicated cysts, while the majority of cysts smaller than 5 cm were asymptomatic.

The proportion of symptomatic cysts also increased with age. The mean age at diagnosis for asymptomatic cysts was 9.1 years (±0.7) (range 0–15.6), significantly younger than for symptomatic cysts: 12.3 years (±0.4) (range 5.5–17.5) ($p < 0.01$). Cysts gradually increased in size with age. However, cyst growth was not constant, as accelerated growth was observed around the age of 12 years (Fig. 1), corresponding to the beginning of puberty.

In this series of 100 patients, 15 patients were under observation and 85 were treated surgically.

The mean age of the patients in the “observational group” was 5.6 years (±2) (range: 0–15.6), and the mean cyst diameter was 33.5 mm (±9) (range: 8–106). Three patients with insufficient follow-up were not included in analysis of the long-term results. The mean follow-up for the remaining 12 patients was 25.4 months (±6). Three patients presented abdominal pain at diagnosis, associated with intracystic hemorrhage in one case. This child's parents refused surgery and he experienced persistent pain for several weeks. The other patients in this group were asymptomatic at diagnosis. All patients in this group had a favorable clinical outcome. The radiological outcome in this group showed complete resolution of the lesion on US imaging for 7 patients (58.3%) after a mean of 29.8 months. The cyst became smaller in the other 5 patients. No significant difference was observed between cyst diameter at diagnosis and the probability of complete cyst resolution, but these results are based on a small sample size.

Cyst was diagnosed prenatally in six patients. It was relatively small at the first postnatal US examination, with a mean diameter of 15.1 mm (±3). None of these patients were symptomatic and all had a favorable outcome with no clinical manifestations. The cyst completely resolved in four patients after a follow-up ranging from 7 months to 4 years and the cyst diameter decreased significantly to 6 mm in two patients.

The mean cyst diameter in the surgery group was 98.1 mm (±5.1) (range: 20–265), and these patients had a mean age of 11.6 years

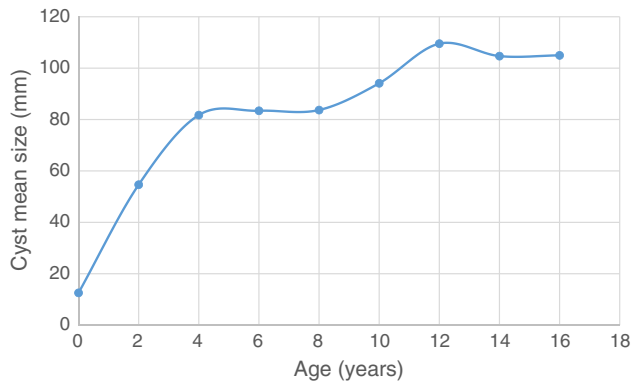

Fig. 1. Course of NPSC diameter with the patient's age.