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CASE REPORT

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Epithelial splenic cysts in children: surgical treatment by cyst-wall "peeling"

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Abstract Primary splenic cysts are a rare finding. Some are large and require surgical removal. The epidermoid type has an epidermal lining, and prevention of recurrence is dependent on complete resection of the cyst wall, preserving the splenic tissue. Several open, laparoscopic, or percutaneous procedures have been proposed with or without splenic resection, but few give completely satisfactory results. Five consecutive splenic epithelial cysts in pediatric patients were treated by parenchyma-sparing complete removal of the cyst wall, which was gently peeled off the splenic tissue without major bleeding in all but one case. Long-term follow-up showed freedom from recurrence.

Keywords Splenic epithelial cyst · Nonparasitic splenic cyst · Surgery · Children

Introduction

Several types of splenic cyst have been described. They are generally divided into parasitic and nonparasitic lesions. Nonparasitic cysts include primary or true cysts, lined by epithelium, and secondary, false, or pseudocysts, which are probably secondary to previous trauma and are much more common (80% of nonparasitic cysts).

True cysts of the spleen are very rare, and are frequently classified as cystic hemangiomas, cystic lymphangiomas, and epidermoid and dermoid cysts. Early appearance in childhood is sometimes reported [6].

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Surgical treatment is optional in small, benign intraparenchymal cysts. Rapid growth and the risk of intracystic bleeding or rupture due to blunt trauma may lead these frequently asymptomatic children to the surgeon [1].

The increased risk of overwhelming postsplenectomy infection in children [5] has prompted organ-saving procedures: partial splenectomy [2, 7]; simple percutaneous aspiration or sclerosis of the cyst [8]; and partial cyst amputation [4]. A significant risk of recurrence is commonly reported for all of them. Most recently, Touloukian et al. [9] proposed partial decapsulation of splenic epithelial cysts (SEC) to preserve splenic function. The cyst wall was almost entirely removed, leaving only a small cuff of lining on the bottom of the residual cyst cavity.

The present report describes five cases of large SECs in pediatric patients. In all cases the entire cyst wall was "peeled" from the splenic parenchyma by an open procedure to exclude the risk of recurrence.

Case reports

Case 1 In a 9-year-old male with a silent left-upper-abdominal mass and a normal hemogram, ultrasound (US) demonstrated a 10-cm cystic lesion of the spleen. Computed tomography (CT) showed a unilocular splenic cyst (Fig. 1). No previous trauma was reported. Percutaneous aspiration was performed through a double "J" drain, which was left in place for 10 days, followed by complete excision of the cyst by smooth dissection of the wall with a small swab and control of bleeding by diathermy and fibrin glue (Fig. 2a–c). One unit of fresh blood was transfused. The excised cyst wall demonstrated an epithelial lining, which confirmed the diagnosis of an epidermoid cyst. The patient was discharged on the 8th postoperative day and is recurrence-free 6 years after the operation.

Case 2 A 6-year-old female was referred for abdominal pain lasting for 3 weeks. US and CT revealed a 6-cm cyst of the spleen. There was no history of trauma; the blood profile and tests for infectious diseases were normal. Complete surgical excision of the cyst wall was performed using the same technique as in case 1. Histology confirmed an epidermoid cyst. The postoperative recovery was uneventful and no recurrence was seen after 4 years.

Case 3 A 14-year-old male was admitted for persistent fever, headache, and vomiting. A history of recurrent abdominal pain

Fig. 1 CT scan showing large, unilocular splenic cyst (20 cm)

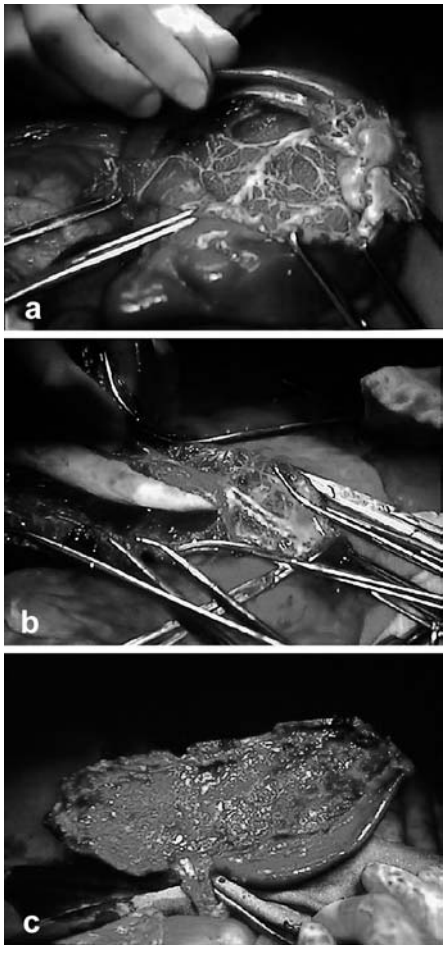

Fig. 2 **a** Inner aspect of splenic cyst. **b** Cyst wall peeled off from the parenchyma. **c** Final appearance of splenic parenchyma

lasting for several months following blunt abdominal trauma (fall from a bicycle) was reported. The physical examination revealed a large spleen and US demonstrated a 15-cm cystic lesion of the upper pole that was confirmed by CT. Adhesions to the diaphragm and left kidney and central displacement of the stomach were present. Complete excision of the cyst, preceded by percutaneous aspiration 1 week previously, was performed. Histology showed squamous epithelial lining. After an uneventful postoperative course he was recurrence-free on follow-up at 2 years.

Case 4 A 9-year-old boy was admitted after blunt abdominal trauma. At US a huge, unilocular splenic cyst was diagnosed (diameter 12 cm) and confirmed by CT. Uncapping of the cyst, leaving a small area of wall strictly adherent to the splenic hilum, was followed by a recurrence 3 years later, which was treated by unsuccessful percutaneous aspiration and sclerosis. At operation, the cyst appeared to have tenacious adhesions to the diaphragm. Complete excision of the cyst wall from the splenic parenchyma required meticulous hemostasis using fibrin glue and hemostatic sponges. The postoperative course was complicated by a left pneumothorax, which required aspiration. No recurrence was detected at 1 year.

Case 5 A 13-year-old male was hospitalized for pneumonia. Investigations for splenomegaly revealed a huge splenic cyst (20 cm diameter) on us. The diagnosis was confirmed by CT. There was no history of previous trauma. After percutaneous pigtail drainage for 3 days, surgical excision of three-fourths of the cyst extrinsic to splenic parenchyma and peeling of the residual wall, which was firmly attached to the splenic hilum, was performed. Postoperative recovery was uneventful. Histology confirmed a SEC. There was no recurrence at 10 months.

Discussion

Five patients with true SECs were treated successfully between February 1994 and April 2001 at the pediatric Surgical Division of S. Camillo Hospital in Rome. In only one case was a single-unit transfusion required. A postoperative drain was left in all cases. Four patients were discharged on the 8th postoperative day; the one who developed a pneumothorax was discharged on the 11th postoperative day. In all cases the definitive treatment, sometimes after a failed attempt at percutaneous aspiration or partial excision, consisted in removing the cyst by peeling it away from the splenic tissue. After follow-up for 10 months to 6 years there were no recurrences. In all cases histology demonstrated the presence of an epidermal lining.

Congenital epidermoid cysts are rare splenic lesions (only 2.5% of all splenic cysts in childhood). Their etiology is still debated: the hypothesis of an embryonic epithelial-cell inclusion from adjacent extra-splenic tissue is opposed by that of a fluid collection secondary to posttraumatic or spontaneous intrasplenic bleeding [3]. Epidermoid cysts have a squamous epithelial lining with intracellular bridges. The trabeculated aspect of the inner cyst wall resembles the chordae tendinae of the cardiac cavities; the content is usually clear, but cloudy fluid with cholesterol crystals, lipids, and erythrocytes may be found.

Indications for surgery are usually based on cyst size and the potential risk of rupture, infection, or intracavitary bleeding. The aim of treatment is to remove the entire cyst, sparing splenic tissue, and avoid recurrence. Several reported procedures (percutaneous, open surgical, laparoscopic) all leave in place remnants of the cyst lining that are potentially responsible for recurrences.

Our five consecutive cases of large (6–20 cm) splenic cysts all had a clear indication for treatment and a high risk of total or partial splenic loss. We adopted a

conservative approach in view of the patients, ages. The epithelial lining could always be peeled off easily without major bleeding, even in deeply-located, large cysts, and the potential risk of recurrence could be thus eliminated.

References

1. Blank E, Campbell JR (1973) Epidermoid cyst of the spleen. Pediatrics 51: 75–84
2. Brown MF, Ross AJ, Bishop HC, et al (1989) Partial splenectomy: the preferred alternative for the treatment of splenic cysts. J Pediatr Surg 24: 694–696
3. Burrig K (1988) Epithelial (true) splenic cysts: pathogenesis of the mesothelial and so-called epidermoid cyst of the spleen. Am J Surg Pathol 12: 275–281
4. Domini M, Aquino A, Rossi C, Pappalepore N, Valentini M, Lelli Chiesa P (1996) Cisti splenica gigante: trattamento chirurgico mediante decapsulazione parziale. RICP 38: 68–71
5. Eraklis AJ, Kerry SV, Diamond LK (1967) Hazard of overwhelming infection after splenectomy in children. N Engl J Med 276: 1225–1229
6. Griscom NT, Hargreaves HK, Schwartz MZ, Reddish JM, Colodny AH (1977) Huge splenic cyst in a newborn: comparison with 10 cases in later childhood and adolescence. A J R 129: 889–891
7. Khan A, Bensoussan A, Ouimet A, et al (1986) Partial splenectomy for benign cystic lesions of the spleen. J Pediatr Surg 21: 749–752
8. Moir C, Guttman F, Jequier S, et al (1989) Splenic cysts: aspiration, sclerosis, or resection J Pediatr Surg 24: 646–648
9. Touloukian RJ, Maharaj A, Ghoussoub R, Reyes M (1997) Partial decapsulation of splenic epithelial cysts: studies on etiology and outcome. J Pediatr Surg 32: 272–274