Spleen preserving management of a non-parasitic splenic cyst in pregnancy

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ABSTRACT

Non-parasitic splenic cysts are rare entities. In pregnancy, they are rarer still, with as few as seven cases reported in the literature. There is little consensus regarding the optimal management of this condition in pregnancy. Although small, the theoretical risk of intrapartum splenic rupture is associated with a fetal mortality rate as high as 70%. The authors of at least three case reports advocate total splenectomy as first-line management of splenic cyst in pregnancy. Paradoxically, spleen conserving surgery is the recognised gold standard treatment for symptomatic splenic cysts in non-pregnant patients. We present a case of a large maternal splenic cyst that was treated successfully with a laparoscopic cystectomy.

KEYWORDS

Splenic cyst – Non-parasitic splenic cyst – Maternal splenic cyst – Pregnancy – Minimally invasive

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There are several published classifications of non-parasitic splenic cysts (NPSCs). The most basic distinguishes between true (primary) and false (secondary) cysts based on the presence of an epithelial lining.^{1,2} Trauma is widely regarded as a common cause of secondary cysts. However, a study from 2012 re-evaluating the histopathology of 540 NPSCs demonstrated that there is no clinically proven evidence that trauma does play a role in their pathogenesis.⁵ While computed tomography (CT) is the investigation of choice, ultrasonography is important in differentiating between non-parasitic (unilocular) and parasitic (multilocular) cysts. Ultrasonography is also a key imaging modality in maternal and paediatric cases, where radiation exposure should be minimised. Malignancy is an important differential diagnosis for splenic lesions and in patients for whom imaging is suggestive of this, fresh frozen biopsies for histological diagnosis and staging are required.

Case history

A 29-year-old woman presented with right loin pain, vomiting, dysuria and urinary frequency. She had a past medical history of recurrent urinary tract infection (UTI) and recent minor trauma involving a fall from a ladder. On clinical examination, she was tender in the right hypochondrium. Urine dipstick testing was suggestive of a UTI.

A diagnosis of pyelonephritis was investigated using abdominal ultrasonography, leading to an incidental finding

of a large perisplenic collection 15cm in maximal diameter with echogenic and avascular features. The spleen itself appeared normal, measuring 10.7cm in length, with no focal parenchymal abnormality. There were no features to suggest that the lesion could be malignant. Computed tomography (CT) confirmed the presence of a large type 1 splenic cyst (Losanoff classification)⁴ and also demonstrated patchy enhancement of the right renal parenchyma, suggestive of multifocal pyelonephritis (Fig 1). Given the short time span between the patient's fall and diagnosis of the cyst, it was suspected that this case most likely represented a primary congenital cyst.

The patient was treated for pyelonephritis and booked for outpatient review. At the follow-up appointment, she was pregnant, prompting review of the initial plan for conservative monitoring. Optimal management was subsequently discussed in a multidisciplinary team meeting. In light of the extremely high risk posed by cyst rupture during pregnancy, cyst drainage and removal was deemed appropriate.

The operation was scheduled during the patient's second trimester to avoid potential disruption of fetal organogenesis. Surgery was performed laparoscopically (Hasson technique). The spleen was identified and the cyst de-roofed. Approximately 1,500ml of dark serosanguinous fluid was aspirated. Splenic adhesions to the anterior abdominal wall and diaphragm were divided. Subsequent de-roofing of the cyst with resection of the medial capsule (Fig 2) was performed using the Harmonic® scalpel (Ethicon, Somerville,

Figure 1 Computed tomography demonstrating a large splenic cyst

NJ, US). Resected tissue was placed in an endobag and removed through the port site. A 500ml peritoneal washout was performed with warmed 0.9% saline. The patient recovered uneventfully and was discharged on the second postoperative day. Histological analysis of the cyst wall demonstrated appearances compatible with a primary splenic epithelial cyst.

Discussion

Non-maternal NPSC

Consensus management of asymptomatic, non-maternal NPSCs is lacking. Some authors advocate surgical management of asymptomatic splenic cysts based on cyst size, suggesting that cysts <4cm resolve spontaneously and those >5cm have a high risk of rupture/infection. In contrast, other studies have shown this threshold to be arbitrary, recommending monitoring of asymptomatic splenic cysts regardless of size.⁵ For symptomatic, non-maternal NPSCs, the general consensus advocates surgical intervention. Surgical options range from percutaneous drainage to splenectomy. Although percutaneous drainage is minimally invasive, it is associated with a high rate of recurrence, infection and bleeding. Conversely, while definitive, total splenectomy exposes the patient to lifelong immunodeficiency and a greater risk of

haemorrhage.⁶ For these reasons, various spleen conserving procedures such as laparoscopic cyst excision have been suggested.

The main concern with laparoscopic cyst excision alone is the potential for recurrence. Two studies measuring the rate of recurrence in paediatric cohorts demonstrated recurrence rates of 64% and 78% respectively.^{4,7} In contrast, a study of adults demonstrated that 5 of 6 cases of NPSCs showed no evidence of recurrence at 28 months.⁸ This discrepancy may reflect a proliferative phase of congenital cysts, which predominates during childhood. A future study directly comparing the recurrence rates of splenic cysts in children versus adults would provide clarification. Attempts to reduce the risk of recurrence including coagulation of the splenic surface with argon laser beams and packing with omentum have been deemed ineffective.¹

Another limitation is that isolated cyst excision may not be suitable for intraparenchymal cysts. Laparoscopic cyst excision with partial splenectomy allows reduced risk of recurrence and preservation of splenic function.^{9,10} An alternative is a two-stage approach, where initial laparoscopic cyst excision is followed by partial splenectomy in the event of recurrence.¹¹ Ambiguity persists as to whether partial resection of the spleen parenchyma is necessary or appropriate.

Maternal NPSC

There is less consensus on the management of maternal NPSCs. The theoretical risk of spontaneous rupture in pregnancy is low; one study attributed 4.5% of cyst ruptures to pregnancy.¹² However, in the event of rupture, 60% occur during the third trimester, resulting in a fetal mortality rate as high as 70%.^{13,14} For this reason, second trimester surgical intervention is universally recommended.

The risks of surgery during pregnancy are mainly associated with general anaesthesia, including spontaneous abortion and premature labour. In laparoscopic methods, there is a risk to the fetus from trocar insertion and carbon dioxide insufflation. These risks must be weighed against the risk of cyst rupture. Minimally invasive approaches such as percutaneous drainage are associated with an unacceptably high complication and failure rate in maternal NPSCs, and are therefore not advocated.⁵ Perhaps surprisingly, three reports on maternal NPSCs suggest laparoscopic total splenectomy as safe practice.¹⁵⁻¹⁷ This contradicts the consensus view of using spleen preserving surgery for non-maternal NPSCs. The present case adds to the evidence that exemplifies the safe and effective use of laparoscopic cyst excision alone in the management of maternal cases. This approach preserves splenic function as well as reducing the risk of haemorrhage and infection associated with resection.

Conclusions

While there is general consensus for spleen preserving surgery in the management of non-maternal splenic cysts, a significant proportion of the available literature advocates total splenectomy in the management of maternal splenic cysts. This dichotomy is almost certainly due in part to the limited number of reported cases. The present case report

Figure 2 The cyst with visible haematoma (A), cyst opening and drainage (B), removal of adhesions (C), cystectomy (D) and final resected specimen (E)

demonstrates the effective use of minimally invasive spleen preserving surgery for treatment of splenic cysts in pregnancy. A two-stage approach should be considered as a risk minimisation strategy for maternal splenic cysts, comprising initial laparoscopic cyst excision followed by monitoring and laparoscopic partial splenectomy in the event of recurrence.

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