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Laparoscopic Partial Cystectomy for Non-Parasitic Splenic Cyst: A Case and Literature Review

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Nonparasitic splenic cysts are rare and no treatment guidelines exists. Laparoscopic partial cystectomy is an attractive minimally invasive spleen preserving treatment option. The aim of this paper is to share our experience and review evidence to support laparoscopic partial cystectomy. A search was performed of PubMed and Ovid Embase. All human studies published in English language describing laparoscopic management of non-parasitic splenic cysts without any age restrictions were reviewed. Forty two studies fulfilled inclusion and exclusion criteria out of 254 studies. The total number of patients were 134. All studies were case reports or retrospective case series which ranged in size from one to 14 patients. Ten studies (24%) involved paediatric patients. Majority of patients were female. Median age ranged from 8.5-43 and median operative time varied between 56-120 min. One study reported bleeding from the cyst wall requiring splenectomy. All other studies did not report any major intra operative or post-operative complications. Median follow up period varied from 6-60 months. Overall recurrence rates varied from 0-100%. When all 134 partial cyst excisions are considered overall, 25 patients developed recurrent cysts leading to a recurrence rate of 19%. Only 13 patients (9.7%) developed symptomatic recurrences needing further intervention. Laparoscopic partial cyst excision is a relatively straightforward safe and effective spleen preserving surgical option with acceptable recurrence rate in the treatment of nonparasitic symptomatic splenic cysts.

Key words: Spleen, cyst, laparoscopy, nonparasitic, excision

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INTRODUCTION

Although accurate information on the incidence is unknown, benign splenic cysts are thought to be an uncommon entity and around 1000 cases have been described. Its incidence seems to be increasing as a result of more frequent use of imaging modalities such as ultrasound and computed tomography. The differential diagnosis for splenic cysts includes cystic neoplasms, benign cysts and abscess. Benign cysts are subdivided into parasitic and nonparasitic. The former occur most commonly throughout Africa, Central America and areas of endemic hydatid disease (Akkoyun *et al.*, 2011). In contrast nonparasitic cysts are common in Europe and North America (Ito *et al.*, 2002; Hansen and Moller, 2004). Non Parasitic Splenic Cysts (NPSC) are further divided into congenital epithelial cysts and pseudocysts (Fowle, 1953; Martin, 1958). Most reports regard pseudo cysts to be the majority of NPSC. However Morgenstern who studied 23 patients with NPSC over 28 years suggest that most NPSC to be congenital based on histopathology along with immune staining as well as macroscopic features of the cyst wall. Proposed a new classification based on the true pathogenesis of cyst dividing non-parasitic splenic cysts as congenital, neoplastic, traumatic and degenerative (Morgenstern, 2002). A recent review of NPSC in children support that congenital cyst are the most common variety in NPSC (Sinha and Agrawala, 2011).

With the understanding of the important immunologic role of the spleen and the importance of prevention of overwhelming sepsis after splenectomy most surgeon now favour spleen preserving surgery (Bisharat *et al.*, 2001; Lynch and Kapila, 1996). As the benefits of minimally invasive surgery is well proven laparoscopic approach would be the optimal option in spleen preserving surgery (Morgenstern, 2002; Comitolo, 2001; Dawes and Malangoni, 1986).

The aim of this study is to present a case of large NPSC treated successfully with laparoscopic partial cystectomy and to review the literature in support of laparoscopic partial cystectomy for symptomatic NPSC.

Case review: Thirty 30 year old female was referred as an emergency due to ultra sound diagnosis of splenic cyst which was associated with 3 months history of gradually increasing left upper quadrant pain, discomfort in the epigastrium and early satiety. She was otherwise healthy with no allergies and was not on regular medications. There was no history of close contact with animals.

Physical examination was unremarkable and contrast enhanced CT scan of the abdomen and pelvis revealed large unilocular splenic cyst measuring 16×13×17 cm without septation and wall enhancement situated in the anterior and superior portion of the spleen (Fig. 1). Her blood tests including anti-echinococcal serology were normal apart from raised CA 19.9 of 244. Non-parasitic splenic cyst was diagnosed. As this was symptomatic and large the surgical options of splenectomy and laparoscopic partial cyst excision were discussed.

The surgical procedure is briefly described and illustrated (Fig. 2). Patient was placed at 45 right lateral position. Pneumoperitoneum was established with 10 mm optical trocar. Twelve millimeter Supra umbilical and two 5 mm left upper quadrant ports were then placed. Splenic flexure and the anterior omental attachment to the cyst were mobilised. Cyst wall was decompressed with aspiration needle and then a small window was created to aspirate about 800 mL of blood stained fluid. The cyst wall and the spleen were freed of adhesions to the diaphragm. The cyst wall was then excised using Harmonic Ace®+7 as close as possible to the splenic tissue. The cyst wall attached to the splenic tissue was left behind and excised cyst wall was extracted through the



Fig. 1(a-b): Pre-operative CT Images showing unilocular cyst compressing the stomach and atrophied splenic parenchyma

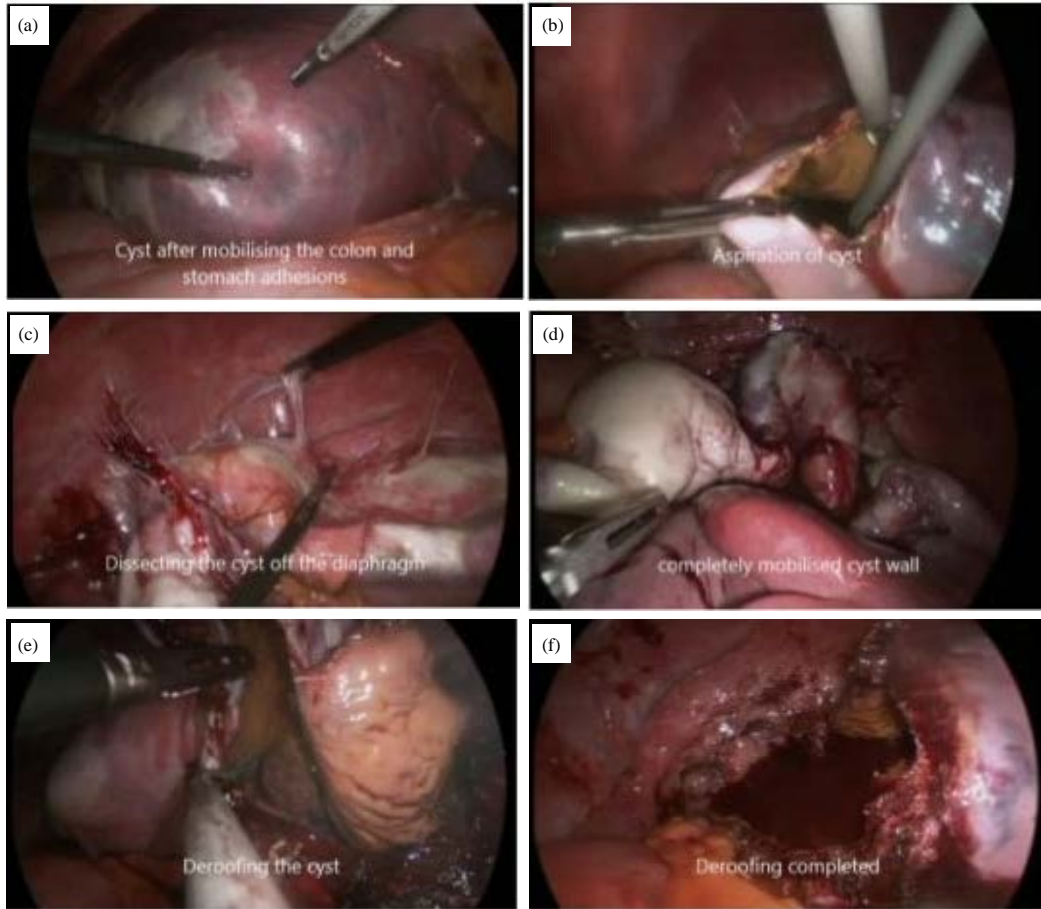


Fig. 2(a-f): Stages of the laparoscopic partial cyst excision after mobilizing splenic flexure and stomach adhesions, aspiration of the cyst, mobilization of the cyst wall off the diaphragm and partial excision of the cyst

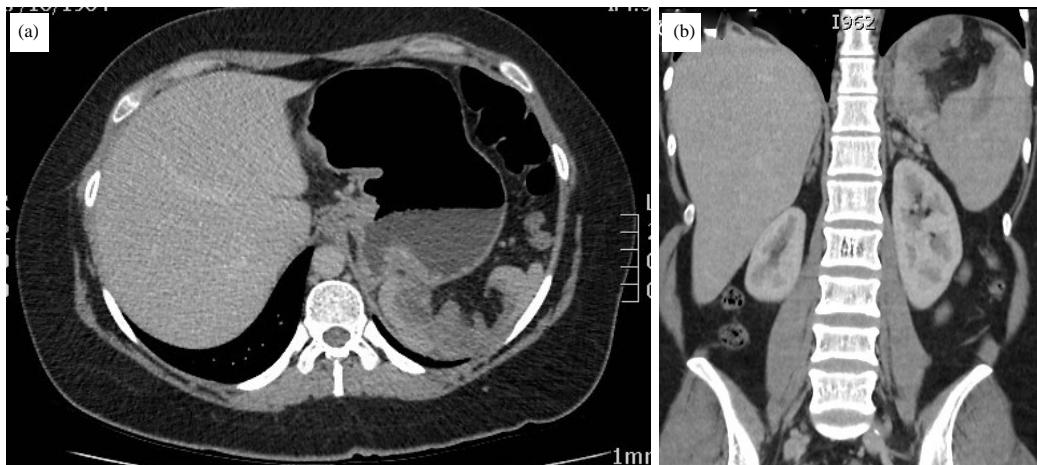


Fig. 3(a-b): Post-operative CT images 3 months after laparoscopic partial cyst excision

umbilical port site. Splenic bed was washed out with saline and drain was placed. Patient had uneventful recovery and discharged after 72 h. Histopathology confirmed congenital epidermoid cyst.

Three months after the procedure follow up CT scan showed excellent results with minor cystic areas. The patient was asymptomatic (Fig. 3). Patient would be followed up regularly with US imaging.

Literature review: PubMed (1950 to September, 2015) and Ovid Embase (1974 to September, 2015), were electronically searched for human studies published in English language describing laparoscopic management of non-parasitic splenic cysts without any age restrictions. Search terms used were laparoscopy, spleen, splenic, cysts, cyst. All abstracts were screened to identify studies that described laparoscopic unroofing/deroofting/partial cystectomy/decapsulation/marsupialization or fenestration of NPSC. Studies that described parasitic cysts, open procedures, cyst aspiration, total or partial splenectomy, cystectomy or total decapsulation were excluded. If laparoscopic fenestration was limited to creating a small window in the cyst wall these were also excluded. Further studies were identified by cross-referencing from relevant articles and abstracts.

RESULTS

Initial literature search revealed 254 articles. After exclusion of the studies that did not meet inclusion and exclusion criteria 62 studies were selected for review of full text articles which resulted in 42 studies describing 134 laparoscopic partial cyst excision of NPSC.

All studies were either case reports or retrospective case series. If a study consisted of 5 or more patients they were considered as case series. Ten studies (24%) involved paediatric patients whereas 32 studies (76%) involved adult patients. Five of the paediatric studies (50%) were case series. Largest case series described 14 patients. Six of the adult studies (19%) were case series, the largest comprising 11 patients. Four of the studies were poster presentations at scientific meetings limited to adult case reports.

Of 134 patients, demographic, operative, follow up data and recurrences were identifiable in eight case series consisting of 63 patients (47%). All case series discussed follow up period and recurrences consisting of 89 patients (66%). Majority of patients were female. Median age ranged from 8.5-43. Median operative time varied between 56-120 min. None of the studies showed intra operative or post-operative complications except in one case series where one case of massive bleeding occurred requiring transfusion and splenectomy. Energy devices used to excise the cyst wall were monopolar diathermy, harmonic scalpel or Ligasure™. Some studies described the use of laparoscopic stapling device.

Median follow up period of the studies varied from 6-60 months. Overall recurrence rates in individual case reports and case series varied from 0-100%. However when the case report of 2 patients with 100% recurrence was excluded then the overall recurrence rate dropped to 0-77%. When asymptomatic recurrences were excluded symptomatic recurrence rates would be 0-44%. When all 134 partial cyst excisions were considered overall, symptomatic and asymptomatic recurrences were 19% (25 patients), 9% (12 patients) and 9.7% (13 patients), respectively.

DISCUSSION

Management of symptomatic splenic cysts lacks clear evidence-based guidelines as a result of its low incidence (Chin *et al.*, 2007). Treatment of choice for NPSC in the past was open splenectomy. The spleen is well visualized laparoscopically. The avoidance of an upper abdominal incision decreases the postoperative pain and discomfort for the patient and shortens length of hospitalization. It is cosmetically more appealing than laparotomy wound. With the development and advancement in laparoscopic technology and skills most surgeons now favour laparoscopic approach in the surgical treatment of splenic diseases (Chin *et al.*, 2007; Mezquita *et al.*, 2007).

Spleen preserving surgery is the optimal treatment in the treatment of NPSC. These options include partial splenectomy, complete excision of the cyst, aspiration of the cyst and partial excision of the cyst. Partial splenectomy and complete excision of the cyst are technically more demanding and associated with increased blood loss. Aspiration of the cyst is associated with higher recurrences. Better results have been reported with aspiration and sclerotherapy. This appears to be safe but require multiple therapy sessions (Rifai *et al.*, 2013).

The first laparoscopic partial cyst wall excision was described by Salky *et al.* (1985). This group decompressed a splenic cyst via a laparoscopic trocar puncture followed by creation of a cyst-peritoneal window. There was no recurrence after 5 years of follow up. Several other studies have shown that laparoscopic partial cystectomy to be safe and effective (Till and Schaarschmidt, 2004; Mackenzie *et al.*, 2004; Dan *et al.*, 2010; Sellers and Starker, 1997; Gianom *et al.*, 2003; Mertens *et al.*, 2007). Results of our review support that laparoscopic partial cyst excision is safe and effective spleen preserving option in the treatment of NPSC. No major complications were reported in our review except in one case out of 134 cyst excisions. Laparoscopic partial cyst excision is technically easier to perform than a total cystectomy and there is minimal blood loss due to the presence of a margin of splenic cortex at the site of cyst excision. Haemostasis is easily achieved with the use of any form of energy device on the cut surface or use of laparoscopic stapling device.

However, argument against this technique is the recurrence rate. The symptomatic recurrence rate in this review varies between 0 and 44%. A European multicentre case series over 25 year period treating 50 children with NPSC consisted of 7 children managed by partial cyst excision. They had a median operating time of 90 min and 12 months median follow up resulting in 44% recurrence (Czauderna *et al.*, 2006). Two other large case series with 13 and 14 patients described symptomatic recurrence rates of 7.6 and 7.1% respectively (Langer *et al.*, 2008; Schier *et al.*, 2007). Two other adult series consisting of 6 and 7 patients reported recurrence rates of 16 and 28%, respectively (Mertens *et al.*, 2007; Chin *et al.*, 2007). Largest adult series involving 11 patients with median operative time of 56 min and median

follow up of 47 months described 0% recurrence rate (Zhang *et al.*, 2007). A recent series consisting of 5 patients with median operative time of 75 min and follow up period of 28 month also reported 0% recurrence rate (Kalogeropoulos *et al.*, 2015). Based on these data and the pooled symptomatic recurrence rates in our review we suggest that recurrence rates would be around 20-25%. In cases of symptomatic recurrence repeat laparoscopic partial cyst excision is a valid alternative depending on the surgeon skill and preference as well as cyst characteristics (Schier *et al.*, 2007).

Indications for treatment of NPSC are presence of symptoms and size of more than 5 cm as these cysts are prone to hemorrhage, rupture and bleeding even when they are asymptomatic. However there is no evidence in the literature to support the 5 cm cut off point as an indication for operative intervention. A recent retrospective study further questions the validity of 5 cm cut off point and we believe it is acceptable to observe asymptomatic patients with serial imaging to assess growth rate and to detect occurrence of symptoms (Kenney *et al.*, 2014).

This review has a number of limitations. All available studies were case reports and retrospective case series with smaller numbers. Most studies were limited to case reports with short follow up period. With longer follow up period the recurrence rate could be higher than the results of this review. Additionally, demographic and operative data were not available in all studies. It's also possible that cases associated with major complications were not reported. Exact details of extent of cyst wall excision was not identifiable in all cases and therefore another unanswered question in this review is the optimum extent of cyst wall excision.

There is no consensus on the extent of cyst wall excision in order to reduce the risk of recurrence. This may explain variable success seen in the literature. Cyst recurrence is possible if any cyst lining remains and the cavity opening close up. It is recommended that cyst is resected as close to the splenic tissue as possible and that as much of the cyst wall as possible should be resected (Till and Schaarschmidt, 2004; Mackenzie *et al.*, 2004; Dan *et al.*, 2010).

Laparoscopic partial cyst excision is not suitable for all NPSC. Cyst in the upper pole are considered critical by some authors (Mezquita *et al.*, 2007). But Mackenzie *et al.* (2004) proved that it is possible to treat upper pole cysts by laparoscopic decapsulation successfully (Mackenzie *et al.*, 2004). We have supported, Mackenzie *et al.* (2004) by successfully performing partial decapsulation of a large superior pole cyst. Cysts located in the splenic hilum, those covered completely by the splenic parenchyma and multiple cysts would require partial or total splenectomy.

CONCLUSION

The laparoscopic management of splenic cysts offers the benefits of minimally invasive surgery: minimal postoperative pain, faster recovery, shorter hospital stay and reduced

morbidity. The surgeon should be prepared to offer to preserve the spleen whenever possible. Laparoscopic partial cyst excision is a relatively straightforward safe and effective spleen preserving surgical option in the treatment of NPSC.

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