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ORIGINAL ARTICLE

Retrospective Study

Laparoscopic splenectomy for splenic littoral cell angioma

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Abstract

AIM: To establish the safety and feasibility of laparoscopic splenectomy (LS) for littoral cell angioma (LCA).

METHODS: From September 2003 to November 2013, 27 patients were diagnosed with LCA in our institution. These patients were divided into two groups based on operative procedure: LS (13 cases, Group 1) and open splenectomy (14 cases, Group 2). Data were collected retrospectively by chart review. Comparisons were performed between the two groups in terms of demographic characteristics (sex and age); operative outcomes (operative time, estimated blood loss, transfusion, and conversion); postoperative details (length of postoperative stay and complications); and follow-up outcome.

RESULTS: LS was successfully carried out in all patients except one in Group 1, who required conversion to hand-assisted LS because of perisplenic adhesions. The average operative time for patients in Group 1 was significantly shorter than that in Group 2 (127 ± 34 min *vs* 177 ± 25 min, $P = 0.001$). The average estimated blood loss in Group 1 was significantly lower than in Group 2 (62 ± 48 mL *vs* 138 ± 64 mL, $P < 0.01$). No patient in Group 1 required a blood transfusion, whereas one in Group 2 required a transfusion. Two patients in Group 1 and four in Group 2 suffered from postoperative complications. All the complications were cured by conservative therapy. There were no deaths in our series. All patients were followed up and no recurrence or abdominal metastasis were found.

CONCLUSION: LS for patients with LCA is safe and feasible, with preferable operative outcomes and long-term tumor-free survival.

Key words: Laparoscopy; Minimal invasiveness; Splenic tumor; Splenectomy; Littoral cell angioma

Core tip: Littoral cell angioma (LCA) is a rare splenic tumor. Consequently, there is a paucity of data in the literature on laparoscopic splenectomy (LS) for LCA. We successfully performed LS in 13 patients with LCA. Compared with patients who underwent open splenectomy, patients who underwent LS required

shorter operative time and suffered lower blood loss. No patient had tumor recurrence. LS is safe and feasible in patients with LCA.

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INTRODUCTION

Littoral cell angioma (LCA) is a rare splenic tumor that was first reported by Falk *et al*[1] in 1991. It arises from cells in the red-pulp sinuses and usually presents with anemia, pyrexia or thrombocytopenia with splenomegaly[2]. Clinically, most of the LCAs described in the literature have been benign; however, several reports have described malignant LCA[3]. Two kinds of morphological presentations of LCA are reported, including the more commonly encountered diffuse multiple nodular form and the rare solitary form[4].

Generally, splenic tumors are rare, and it is difficult to establish a definite preoperative diagnosis of whether they are malignant or benign. Splenectomy is generally indicated for patients with splenic tumors because of the possibility that the lesion is malignant[5]. Since laparoscopic splenectomy (LS) was introduced in 1991[6], it has shown advantages over open splenectomy in terms of lower blood loss, shorter postoperative stay and fewer surgery-related complications[7]. However, it is still controversial to perform total LS for a splenic tumor[5]. There is a paucity of data in the literature on LS for LCA because of its rarity.

In this study, we reported the largest series of LS for LCA, aiming to acquire a better understanding of LCA and to establish the safety and feasibility of LS in this setting.

MATERIALS AND METHODS

From September 2003 to May 2013, 27 patients underwent splenectomy and were diagnosed with LCA by postoperative pathological and immunohistological examinations. The data were collected retrospectively by chart review in terms of demographic characteristics, operative details, postoperative details and follow-up outcomes. This study was approved by the Ethics Committee of Sichuan University, Chengdu, China.

Operative procedure

Patients received general anesthesia and were placed in the right semi-decubitus position, with the left side elevated by approximately 60° and the operating table slightly tilted to the reverse Trendelenburg position. The surgical procedure for LS has been described previously[8]. Four ports were used for all patients. A 10-mm trocar was placed at the upper umbilicus for a 10-mm, 30° camera. A 5-mm trocar was placed at the subxiphoid position, and another 5-mm trocar was placed in the left axillary line below the lower pole of the spleen. A 12-mm trocar was placed at the left mid-clavicular line, below the margin of the spleen, for the use of the ultrasonic dissector and linear laparoscopic vascular stapler. We dissected the perisplenic ligaments in the order of splenogastric ligament (including the short gastric arteries), splenophrenic ligament, splenic flexure attachment and splenorenal ligament. After all the attachments and ligaments were entirely dissected, the splenic hilum was transected with a linear laparoscopic vascular stapler (Echelon 60 ENDOPATH Stapler; Ethicon Endo-Surgery, Cincinnati, OH, United States). The spleen was placed in a retrieval bag, morcellated with forceps and retracted *via* a 12-mm incision. A closed suction drain was routinely placed in the splenic fossa. The amylase level of the drainage fluid was measured.

Definitions

Operative time was defined as the time from the first incision to skin closure. The splenic size was defined as the longitudinal diameter. Splenomegaly was defined as longitudinal diameter > 12 cm[9]. Morbidity was defined as any complication associated with the operation within 30 d of surgery. Pancreatic fistula (graded A-C) was defined by the International Study Group on Pancreatic Fistula (ISGPF)[10].

Statistical analysis

Numerical data are expressed as mean ± standard deviation. Statistical analyses were performed using SPSS for Windows version 16.0. Differences between variables were compared using the nonparametric Mann-Whitney *U* test, Student's *t* test, χ^2 test, and Fisher's exact test. $P < 0.05$ was considered statistically significant.

RESULTS

The demographic and clinical characteristics of the patients are shown in Table 1. There were 15 female and 12 male patients, with a female-to-male ratio of 1.25 to 1. The median age was 45 years (range: 7-65 years). There was no significant difference between the two groups in terms of demographic characteristics.

Overall, 12 patients (44.4%) were asymptomatic, with splenic lesions detected by routine physical examination. The most frequent symptom was abdominal pain (33.3%). In terms of radiographical character, only three patients (11.1%) presented with a solitary lesion. A typical computed tomography image is shown in Figure 1. Splenomegaly (18 cases, 66.7%) was a common presentation of LCA, and thrombocytopenia (14 cases, 51.9%) was also frequent. No significant difference was found between

Table 1 Demographic and clinical characteristics *n* (%)

Variables	Group 1	Group 2	*P* value
Cases	13	14	-
Age (yr)	47.2 ± 11.0	43.2 ± 19.2	NS
Sex (M/F)	4/9	8/6	NS
Platelet count (× 10^9/L)	93.3 ± 46.1	103.7 ± 45.9	NS
Clinical symptoms			NS
No symptoms	6 (46.2)	6 (42.8)	
Abdominal pain	5 (38.5)	4 (28.6)	
Hypersplenism	8 (61.5)	6 (42.9)	
Radiographical features			NS
Isolated mass	1 (7.7)	2 (15.4)	
Multiple masses	12 (92.3)	13 (84.6)	
Splenomegaly			NS
Yes	9 (69.2)	9 (64.3)	
No	4 (30.8)	5 (35.7)	
Thrombocytopenia			NS
Yes	8 (61.5)	6 (42.9)	
No	5 (38.5)	8 (57.1)	

Data are numbers with percentages in parentheses unless otherwise indicated. NS: Not significant.

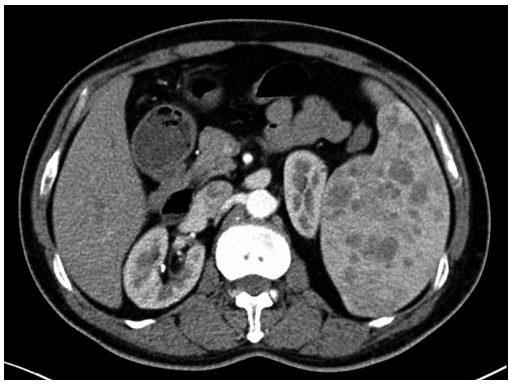

Figure 1 Computed tomography image of a littoral cell angioma reveals multiple splenic lesions and splenomegaly.

the two groups in terms of clinical and radiographical characteristics.

The operative and postoperative details are shown in Table 2. We successfully performed LS for 12 of 13 patients with LCA. One patient required conversion to hand-assisted LS (HALS) because of extensive perisplenic adhesion. The spleen size was comparable between the two groups (17.2 ± 4.8 cm *vs* 16.1 ± 3.9 cm, $P = 0.524$). The mean operative time for patients in Group 1 was 128 ± 37 min (range: 80-230 min). The patients in Group 2 required a significantly longer operative time (177 ± 25 min, range: 150-230 min, $P < 0.001$). The average estimated blood loss for patients in Group 1 was also significantly lower (62 ± 48 mL *vs* 138 ± 64 mL, $P = 0.002$). No patient in Group 1 required blood transfusion, whereas one patient (7.1%) in Group 2 required blood transfusion. There were no deaths in either group.

Six patients in our series suffered from complications, including two (15.4%) in Group 1 and four (28.5%) in Group 2. One patient in Group 1 suffered

Table 2 Operative outcomes and postoperative details *n* (%)

Variables	Group 1	Group 2	*P* value
Operating time (min)	128 ± 37	177 ± 25	< 0.001
Estimated blood loss (mL)	62 ± 48	138 ± 64	0.002
Transfusion	0 (0)	1 (7.1)	NS
Conversion	1 (7.7)	-	-
Spleen size (cm)	17.2 ± 4.8	16.1 ± 3.9	NS
Length of stay (d)	4.9 ± 1.1	7.1 ± 2.4	0.005
Time to oral intake (d)			
Complications	2 (15.4)	4 (28.5)	NS
Incision infection	0 (0)	1 (7.1)	
Pancreatic fistula	1 (7.7)	0 (0)	
Portal vein thrombosis	1 (7.7)	0 (0)	
Pulmonary infection	0 (0)	1 (7.1)	
Abdominal fluid collection	0 (0)	2 (14.3)	

Data are numbers with percentages in parentheses unless otherwise indicated. NS: Not significant.

from Grade A pancreatic fistula, which was diagnosed by routine examination of amylase levels in the drainage fluid. One patient suffered from portal vein thrombosis, which was diagnosed by ultrasonographic examination. Both patients were cured by conservative therapy. Four patients in Group 2 suffered from incision infection, pulmonary infection and abdominal fluid collection. All patients were cured by conservative therapy. There were no deaths in either group.

All patients were followed up *via* outpatient visits and/or telephone interview. The mean follow-up period was 42 mo (range: 9-125 mo). No patient suffered from tumor recurrence or abdominal dissemination. The platelet count returned to normal in 13 of 14 patients.

DISCUSSION

LCA is a rare primary splenic vascular tumor that originates from the littoral cells lining the splenic red pulp sinuses. Although the majority of LCA tumors are benign, there are two subtypes of malignant LCA reported in the literature, including littoral cell angiosarcoma and littoral cell hemangioendothelioma[11]. There are few clinical data regarding laparoscopic management of LCA. To date, our study included the largest series of LS for LCA, which enabled us to establish more definite conclusions regarding the safety and feasibility of LS in this setting.

The clinical presentations of LCA ranges from completely asymptomatic to symptoms such as abdominal pain, splenomegaly and thrombocytopenia[12,13]. Although there is no age predilection, LCA usually occurs in adults and appears to be rare in children. Only two patients (7.4%) who suffered from LCA were children in our series. The majority of LCAs were multiple, although solitary lesions were also reported. It is difficult to establish an accurate diagnosis of LCA preoperatively. At present, the final diagnosis is only possible *via* histopathological examination[14,15]. Fine-needle aspiration is performed for

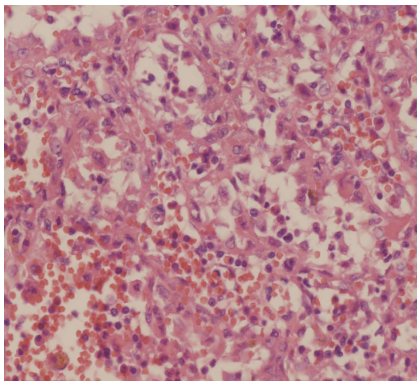

Figure 2 Histopathological presentation of a morcellated spleen with littoral cell angioma.

cytological diagnosis of splenic tumors[16]. However, this procedure is not routinely recommended for a mass in the spleen because of poor specificity, the risk of bleeding and tumor cell dissemination if the tumor is malignant[5].

Splenectomy and long-term follow-up are indicated for LCA because of its malignant potential[17]. Given the lower blood loss, shorter postoperative stay and fewer surgery-related complications, LS has become the gold standard for many hematological disorders, such as immune thrombocytopenic purpura and hemolytic anemia. However, there are just a few case reports regarding LS for LCA[5,18,19]. Rosen *et al*[19]. reported the first case of LS for LCA in 2002. Blansfield *et al*[18] reported the second case of LS for LCA in 2005. Both of those patients underwent successful LS, with favorable operative outcomes and no morbidity. Yano *et al*[5] reported one case of hand-assisted splenectomy for LCA in 2003. That patient was also discharged uneventfully.

However, there are still many concerns about LS in the setting of splenic tumors. LCA may be associated with splenomegaly, even a massive splenomegaly. It is a technical challenge to perform LS for patients with a massive splenomegaly. Furthermore, some surgeons have stated that an intact specimen is necessary for histological examination, and others argue that tumors treated laparoscopically may deteriorate the oncological outcome[20].

In our study, there were 13 patients in the LS group, including nine with splenomegaly. All patients, including those with splenomegaly, underwent successful LS. The mean operating time was 128 min and the mean blood loss was 62 mL, which were comparable with the data in the literature. No patient required conversion to open surgery or blood transfusion. Only one patient converted to HALS because of perisplenic adhesion. The introduction of HALS has enabled surgeons to insert their hands into the abdomen while maintaining the pneumoperitoneum. This technique allows surgeons to recover tactile sensation, enables them to obtain quick access to hemorrhages, and facilitates the retrieval of the spleen from the abdomen. HALS enables surgeons to carry out LS successfully without conversion to open surgery[21]. Overall, it is technically feasible to perform LS for patients with LCA.

Fais *et al*[22] stated that laparoscopic procedures for tumors are not suitable because of high local recurrence after long-term follow-up. They concluded that tumor recurrence might be caused by a pneumoperitoneum. Fortunately, the LCA was located in the splenic parenchyma. The splenic capsule can prevent the dissemination of tumor cells. Subsequently, to prevent tumor cell dissemination, additional attention should be paid to prevent splenic capsule rupture during dissection. We also highlighted the importance of sufficient elevation of the upper pole of the spleen, which is crucial so that the first stapler can cross the splenic hilum. Kawanaka *et al*[21] reported seven cases of uncontrollable bleeding during transection of the splenic hilar pedicles with an endoscopic linear vascular stapler because the first stapler failed to cross the entire splenic hilar pedicles. In our practice, we experienced a patient with hypersplenism caused by liver cirrhosis, which required conversion to open surgery, because of the stapler crossing the splenic parenchyma, caused by insufficient elevation of the upper pole of the spleen. This could be a major problem if we were dealing with splenic tumors, especially malignant tumors.

All our specimens were put into a retrieval bag, morcellated and retrieved *via* the 12-mm incision. During this procedure, additional attention should be paid to keep the retrieval bag intact, which is crucial to prevent tumor cell dissemination. The morcellated specimens did not interfere with the final histological diagnosis of LCA (Figure 2). However, if an intact specimen is required, an additional incision is required for retrieval. All patients were followed up and no patient suffered from tumor cell implantation in the trocar incisions or abdominal dissemination.

There were several limitations associated with our study. The study was retrospective. The sample size was relatively small, which precluded us from establishing a definite conclusion regarding the safety and feasibility of LS in the setting of LCA. Thus, multicenter prospective studies are required to achieve a better understanding of LCA and identify the safety and feasibility of LS in this setting.

In conclusion, LCA is a rare splenic neoplasm. Splenectomy is an effective therapeutic strategy, with long-term tumor-free survival. It is safe and feasible to perform LS in patients with LCA. However, LS for LCA should be performed by experienced laparoscopic surgeons and additional attention should be paid to prevent tumor cell dissemination.

COMMENTS

Background

Littoral cell angioma (LCA) is a rare splenic neoplasm. Consequently, there is a paucity of data in the literature on laparoscopic splenectomy (LS) for LCA.

Research frontiers

The authors included the largest number of LCAs reported in the literature and

presented the demographic characteristics, clinical presentation, radiological characteristics and long-term outcomes of splenectomy for LCA.

Innovations and breakthroughs

This is the first study concerning the safety and feasibility of LS in patients with LCA. Compared with open splenectomy, patients who underwent LS required shorter operative time and had lower blood loss. Patients who underwent LS suffered from fewer complications and required a shorter postoperative stay. The patients in the two groups had comparable long-term tumor-free survival rates.

Applications

LS is safe and feasible for patients with LCA. However, LS for LCA should be performed by experienced laparoscopic surgeons and additional attention should be paid to prevent tumor cell dissemination.

Terminology

LCA is a rare splenic tumor that arises from cells in the red-pulp sinuses of the spleen.

Peer-review

Although a retrospective small case series, it was a well-conducted study concerning a rare splenic tumor, approached laparoscopically. The authors describe the feasibility and usefulness of LS for splenic LCA.

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