

## Laparoscopic Treatment of Intraabdominal Cystic Lymphangioma

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### Rezumat

#### *Tratamentul laparoscopic al limfangioamelor chistice intraabdominale*

Limfangiomul chistic (CL) abdominal la adulți este o tumoră benignă rară a sistemului limfatic.

*Material și metodă:* Am efectuat un studiu retrospectiv din ianuarie 2002 până în decembrie 2014 referitor la 18 pacienți care au suferit ablația chirurgicală a unui CL abdominal, din care 9 pacienți cu abord laparoscopic. La pacienții cu abord laparoscopic s-au urmărit localizarea, mărimea și numărul, diagnosticul, tratamentul și rezultatele postoperatorii.

*Rezultate:* Au fost 8 femei și 1 bărbat cu o vârstă medie de 35,6 ani (cu extreme 20-51 ani). Clinic, principalul simptom a fost o durerea abdominală întâlnită la 8 pacienți (88,8%). Examenul clinic a relevat o tumoră abdominală la 5 pacienți (55,5%). CL a fost asimptomatic la patru pacienți; descoperirea CL a fost efectuată preoperator imagic ecografic pentru o altă patologie (n = 3) sau intraoperator (n = 1). Ecografia și CT permit de obicei, diagnosticul preoperator. Cea mai frecventă localizare a fost în mezenter (n = 3; 33%) și în spațiul retroperitoneal stâng (n = 3; 33%), urmat de spațiul retroperitoneal drept, bursă omentală și mare epiploon câte 1 caz. S-a practicat: chistectomie totală laparoscopică fără puncție chist la doi pacienți și evacuarea chisturilor mari, urmată de chistectomie totală în șapte cazuri. Nu am înregistrat conversii, morbiditate sau mortalitate. Spitalizarea postoperatorie a fost de 3,4 zile. Nu am

înregistrat recidive după o urmărire medie 28 de luni.

*Concluzii:* Abordul laparoscopic este gold standard în tratamentul CL intraabdominale. Recomandăm excizia completă chirurgicală pentru a evita recidiva.

**Cuvinte cheie:** limfangioame chistice, cistectomie laparoscopică, tumori chistice retroperitoneale

### Abstract

The abdominal cystic lymphangioma (CL) in adults is a rare benign tumor of the lymphatic system.

*Methods:* We report a retrospective study from January 2002 to Decemberr 2014 concerning 18 patients who underwent surgical removal of a CL, 9 patients with laparoscopic approach included. The localization, size, and number, diagnostic, treatment and results have been reported for patients approached laparoscopically.

*Results:* There were 8 women and 1 man with median age at diagnosis was 35,6 years (range 20–51 years). Clinically, the main symptom was an abdominal pain found in 8 patients (88.8%). Physical examination revealed an abdominal mass in 5 patients (55.5%). The CL was asymptomatic in four patients; the discovery of CL was performed preoperatively during an ultrasound for another pathology (n=3) or intraoperatively (n=1). US exam & CT scan usually allow the preoperative diagnosis. The most common site was shared equally between the mesentery (n = 3; 33%) and left retroperitoneum (n = 3;33%), followed by the right retroperitoneum and the posterior cavity of the lesser omentum and great omentum, each one case. The most common procedures performed were: laparoscopic total cystectomy of a closed cyst in two patients and

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evacuation of larger cysts followed by total cystectomy in seven patients. No conversion, no mortalities and no morbidity was noted. Mean hospital stay was 3.4 days. No recidive after 28 months in the average after treatment.

**Conclusions:** The laparoscopic approach is the gold standard in the treatment of intraabdominal CL. We recommend complete surgical excision to avoid recurrence.

**Key words:** cystic lymphangioma, laparoscopic cyctectomy, retroperitoneum cystic tumor

## Introduction

Cystic lymphangioma (CL), usually discovered in childhood, is a benign rare malformation of lymphatic vessels, affecting mostly the cranio-facial and cervical region (75%), chest and axilla (20%) and abdominal cavity (5%)(1).

Half of these lesions would be present at birth, where as 90% of cystic lymphangiomas would grow until the age of 2 (2). Its discovery in adults is rare. The common forms found in adults are mesenteric and/or retroperitoneal. CL represents 7% of abdominal cystic lesions in adults. The prevalence of this tumor is as rare as 1 in 20,000 to 250,000 people (2).

The aims of this study is to clarify the clinical manifestations of this disease, revealing the diagnostic difficulties and the therapeutic problems.

## Methods

A retrospective study of adult patients operated on for abdominal CL in First Surgical Clinic Iasi between 2002 and 2014 and Department of Surgery & Oncology, "St. Mary" Clinic, Cambrai, France.

Age, sex, functional signs, physical and radiological findings of each patient were documented. All patients had an ultrasound/or Abdominal CT scan performed. The number, localization and size of the tumor were noted.

Surgical treatment by laparoscopic approach consisted of a complete resection of the cysts. The histopathological examination of the resected specimen was made in all cases.

All patients were followed by a physical examination and abdominal ultrasound.

## Results

Out of a total of 18 intra abdominal CL who underwent surgical removal, 9 patients were approached laparoscopically.

There were 8 women and 1 man with a sex ratio equal to 0,1. Median age at diagnosis was 35.6 (range 20–51) years. Median BMI of 25 kg/same (range 23.5 to 32.3).

Clinically, the main symptom was an abdominal pain found in 8 patients (88.8%). Other signs were: subocclusive syndrome (n=1) and dysuria (n=1). Physical examination revealed an abdominal mass in 5 patients (55.5%). The CL was

asymptomatic in four patients; the discovery of CL was performed preoperatively during an ultrasound for another pathology (n=3) or intraoperatively (n=1). (Table 1).

All cases were examined preoperatively by abdominal ultrasound and 7 by CT scan and MRI (two cases). Five patients were diagnosed preoperatively by abdominal ultrasound or/and abdominal CT scan. By imaging: – were noted the number, location and size of cysts. (Fig. 1, 2).

The most common site was shared equally between the mesentery (n = 3; 33%) and left retroperitoneum (n = 3;33%), followed by the right retroperitoneum and the posterior cavity of the lesser omentum and great omentum. Omental lymphangiomas are rare intra-abdominal tumors (3). We describe a case of omental lymphangioma. The lesion was correctly diagnosed intraoperatively and complete excision of the cyst was performed laparoscopically.

These intra-abdominal cystic lesions were unilocular (n = 8), multilocular (n = 1). As related to the anatomical features of the cysts, their size ranged from 8 to 20 cm with a median size of 11.4 cm.

The initial diagnosis varied from a CL (n = 4), a hydatid cyst (n = 1), a cyst of the mesentery (n = 1), ovarian cyst (1), pancreatic cyst (2). (Table 1)

Surgical treatment consisted of a complete resection of 9 cysts. Laparoscopic procedure: operating room set-up and trocars placement according the CL site; Umbilical approach to create CO2 pneumoperitoneum: 22.22% (N=2) open approach, 77.78% (N=7) Veress needle. Resection in contact with CL using monopolar, bipolar and Ligasure instruments. The most common procedures performed were: total cystectomy of a closed cyst in 2 patients and evacuation of larger cysts followed by total cystectomy in seven patients. (Fig. 1, 2)

There were no mortalities and morbidity postoperatively. Mean hospital stay was 3.4 days.

In the long-term, all patients were followed up and monitored by a physical examination and abdominal ultrasound: no relapse 28 months in the average after the treatment.

The histopathological examination of the resected specimen was consistent with typical findings of a CL: dilated lymphatic vessels, lined with flattened endothelial cells without atypia, and with abundant lymphoid tissue. In addition, smooth muscle cells and foam cells containing lipid material were reflected in their wall (Fig. 3). The diagnosis was specified by immunohistochemistry (Fig. 4).

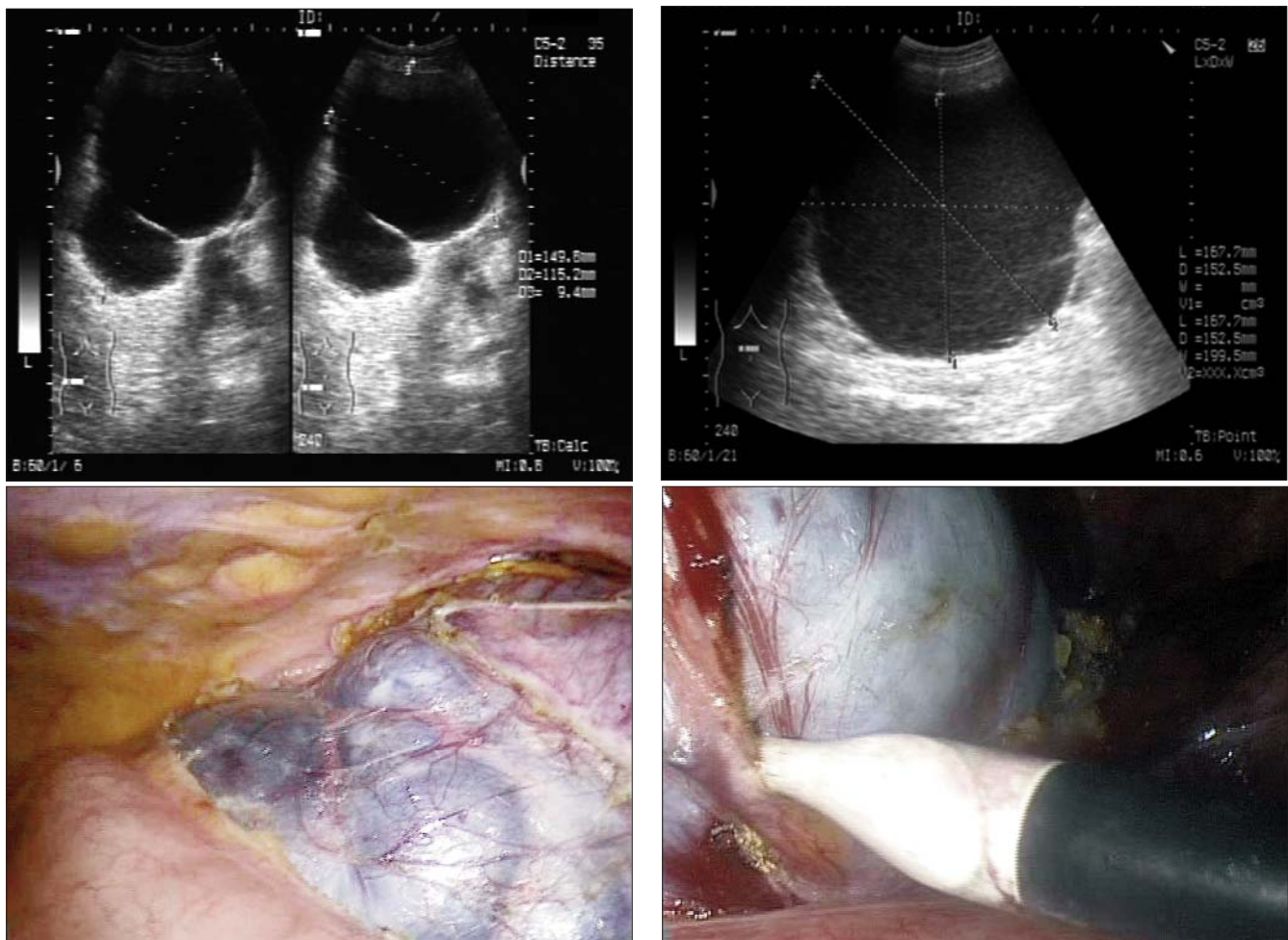
## Discussions

The most likely hypothesis for the development of a CL is a congenital anomaly (2). It is accepted that the peripheral lymphatic system develops from primitive bags from the venous system. During embryogenesis, a faulty connection between lymphatic channels and venous system causes formation of lymphatic cyst was identified (2-4).

The intra-abdominal lymphangiomas are essentially located in the mesentery, as was 3 cases in our series, but may relate to the gastrointestinal tract, spleen, liver, kidneys, adrenals, and pancreas (5). Retroperitoneal location appears to be rare

**Table 1.** Characteristic dates of the patients

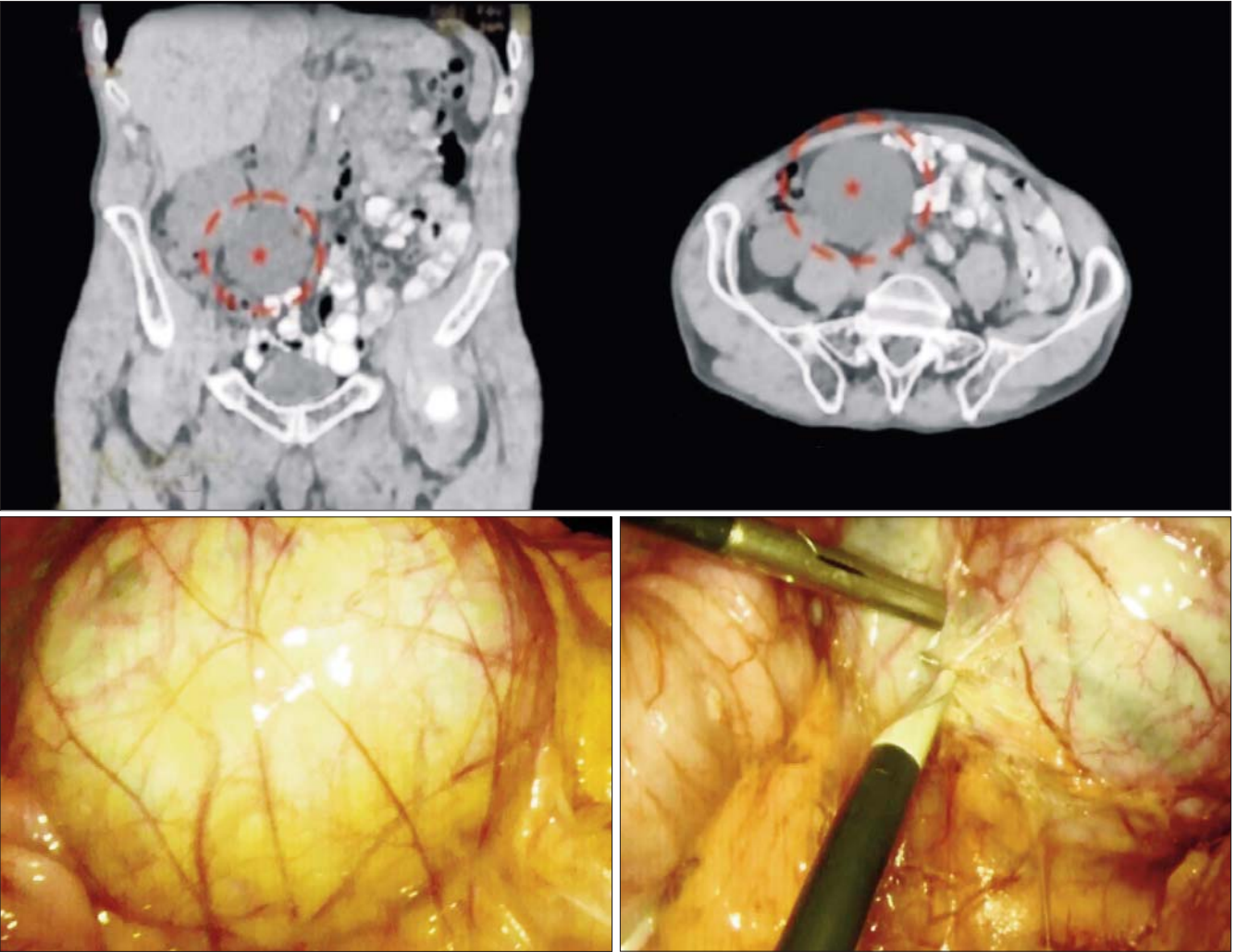
Case No	Sex	Age	Signs	Preoperative diagnosis	Site	Size (cm)
1	M	51	Abdominal pain + mass	Pancreatic cyst	Mesentery	8
2	F	32	Abdominal pain + mass	CL	Mesentery	18
3	F	30	Abdominal pain + mass	CL	Retroperitoneal	10
4	F	45	-	CL	Retroperitoneal	12
5	F	20	Abdominal pain	Ovarian cyst	Omentum	12
6	F	73	-	CL	Retroperitoneal	12
7	F	55	Abdominal pain	Ovarian cyst	Retroperitoneal	6
8	F	47	Abdominal pain	CL	Retroperitoneal	7
9	B	50	-	Adenopathy	Retroperitoneal	6



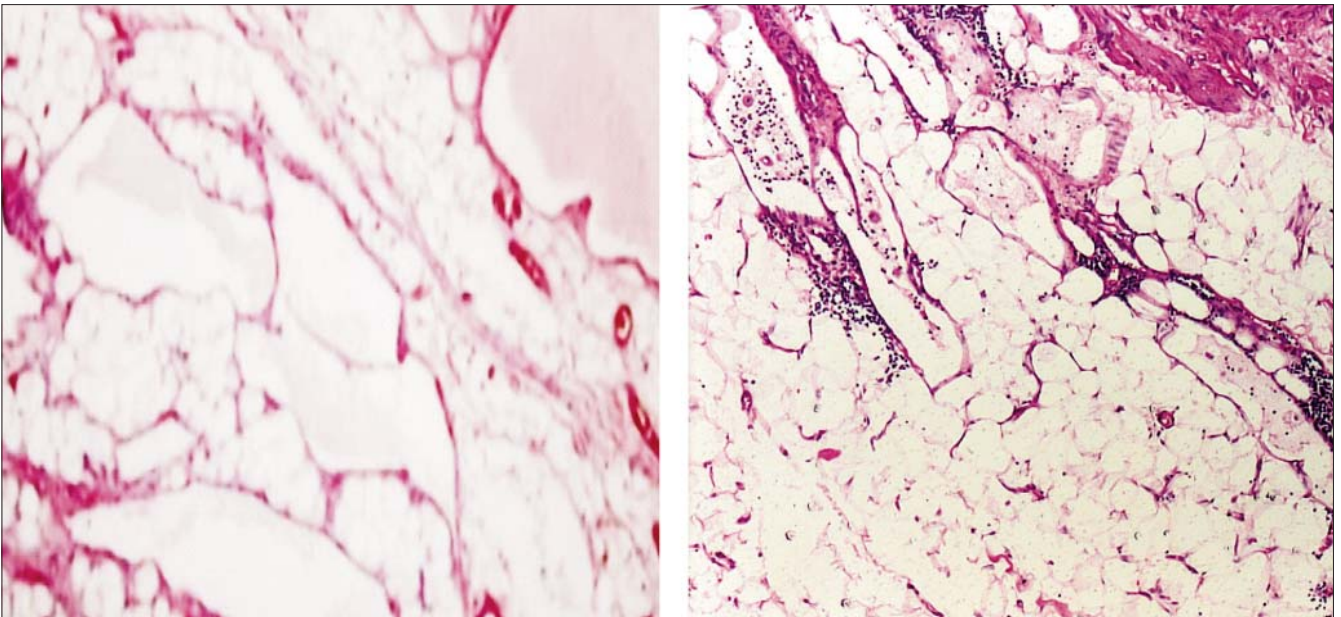
**Figure 1.** Cystic mass located in retroperitoneal space: ultrasound (167/ 12/ 149 mm hypoechogenicmass, 1000 cm3) and intraoperative view of total cystectomy

according to some authors in contrast to our results (33%). The CL are vascular malformations and tumors for which there has never been demonstrated a malignant potential (2).

In the literature, men and women are affected similarly in adulthood, whereas our results show a marked female predominance (6)

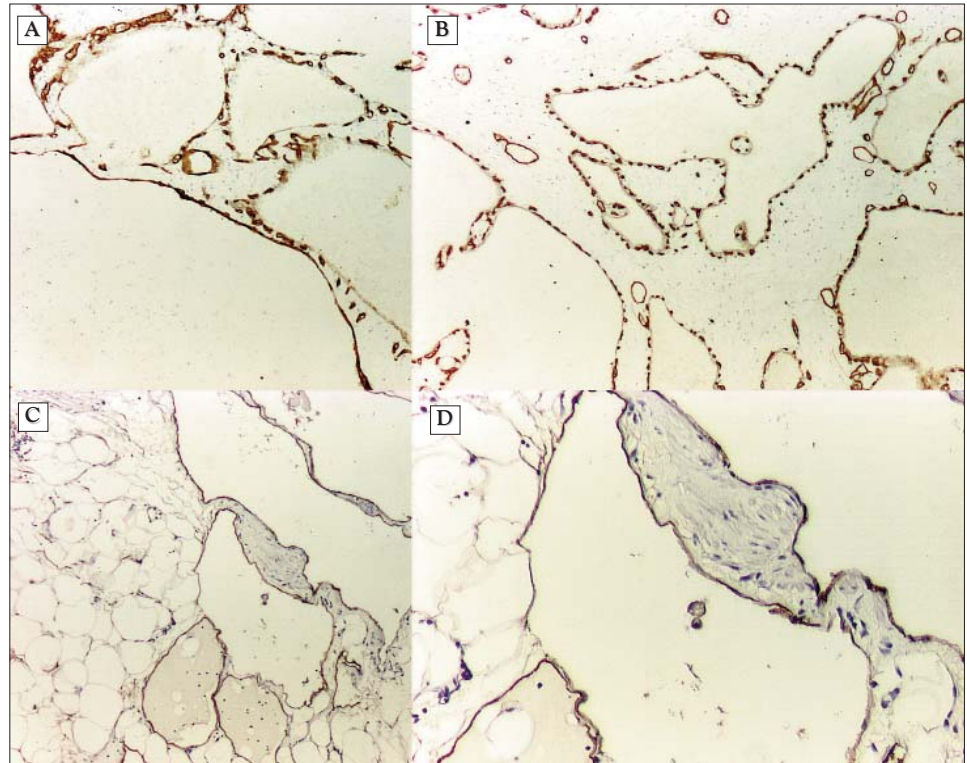


**Figure 2.** Unilocular cystic mass located in mesentericocolic space: CT scan and intraoperative view of total cystectomy



**Figure 3.** Histopathological exam: numerous dilated lymphatic vessels, of varying size and a few disorganized bundles of smooth muscle

**Figure 4.** Immunohistochemistry: (A) CD31 positive diffuse in vascular endothelium x4; (B) CD34 positive diffuse x 4; (C,D) D2-40 positive diffuse in lymphatic vessels x 4



Many lymphangiomas are asymptomatic. The clinical manifestations of abdominal cystic lymphangioma are highly polymorphic. A large tumor volume usually causes abdominal pain, (the most common symptom - 88% in our series) and an increase in waist circumference, a palpable mass (66%), intestinal obstruction, or a volvulus. Other complications can cause acute clinical presentations, such as a hemorrhage cyst, a secondary infection or spontaneous rupture of cysts (2).

For diagnosis, ultrasound examination was useful initially. In the present study, it detected the lesion in 100% of cases (cyst mass). It typically shows a unilocular cystic mass, thin wall, containing hypoechoic fluid (7). The CT scanner has proved to be an excellent initial diagnostic tool. It usually shows a tumor content homogeneous, hypodense, that does not take the contrast. The presence of sediment at the bottom of the cyst is a sign highly suggestive of the diagnosis of CL (8). MRI – is gold standard. MRI better clarifies the nature of the contents of the cysts.

Diagnosis of intra-abdominal CL lymphangioma is difficult because of its rarity, and because other pathologies may have the same radiological signs. The diagnosis is confirmed by anatomopathological examination with immunohistochemistry. The differential diagnosis includes: lymphomadigestive duplication, - large ovarian cyst, hydatid cysts (in our country - endemic area), mesenteric cyst (malignant potential), pancreatic cyst (2).

The aspiration of the cyst, with or without injection of sclerosing agents (Bleomycin, Tissuecol, Ethibloc), has long-term effects, such as frequent recurrences, up to 100% in some series (8). The complete treatment is surgical excision, in order to prevent recurrence.

Laparoscopic removal of CL has proved to be feasible. This

technique has several advantages over conventional laparotomy, including minimal trauma, reduced pain, and early recovery after surgery.

The laparoscopic approach can be successfully and safely performed by an experienced surgeon. Although the laparoscopic treatment may be feasible and has been reported in the literature, the extension of the mass to the retroperitoneum can make a minimally invasive surgical approach difficult (9). A total cystectomy cyst closed in case of small cyst or cyst puncture-opened after evacuation of cystic contents in case of large cysts. Excision of adjacent organs should be avoided in uncomplicated as there is always a dissection plane (10).

## Conclusions

The abdominal CL in adults is a rare benign tumor of the lymphatic system. For diagnosis, ultrasound examination is useful initially. The CT scanner is an excellent diagnostic tool. MRI better clarifies the nature of the contents of the cysts.

If the patient is asymptomatic, the general rule is to continue monitoring the patient regularly. If symptomatic lesions occur complete surgical excision seems to be the best option to reduce the risk of recurrence. The laparoscopic approach is the feasible gold standard. It certifies the diagnosis and allows complete surgical excision, which reduces the risk of relapse.

## Conflicts of interest

No potential conflict of interest relevant to this article was reported.

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