# **Clinical Analysis of Intraperitoneal Lymphangioma**

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#### Abstract

**Background:** Intraperitoneal lymphangioma (IL) used to be thought of as a benign lymphatic malformation with a low rate of preoperative diagnosis. This retrospective study aimed to explore the connection between the cysts and clinical manifestation and imaging characteristics, and to study diagnostic confusion, therapeutic principles and potential recurrent reasons, to further enhance the comprehension of this rare disease.

Methods: Here, we retrospectively reviewed 21 patients diagnosed with IL. Age, sex, complaints, physical findings, and imaging features of each patient were documented. The therapies, postoperative complications and treatments were discussed.

**Results:** Symptomatology included eight patients (38%) with intermittent dull pain in the abdomen, and three patients (14%) complained of abdominal persistent pain. The physical examination revealed an abdominal mass in 16 patients (76%), and eight (38%) were reported no discomfort. IL was correctly established preoperatively in 19 patients (90%). Patients were treated using laparotomy, except one who was treated with laparoscopy. Two recurrences were noted during follow-up.

**Conclusions:** IL should be suspected in any patient with a mobile abdominal mass and surgery is required immediately after discovery of the tumor.

Key words: Abdomen; Imaging; Lymphangioma; Recurrence; Symptoms; Therapeutic Principles

### INTRODUCTION

Intraperitoneal lymphangioma (IL) is a type of benign cyst with an incidence of approximately 1%.<sup>[1,2]</sup> Lesions often take place in mesentery, retroperitoneum.<sup>[3]</sup> Most previously reported ILs are asymptomatic and are often incidentally found through imaging investigation or during surgery for other unrelated causes.<sup>[4]</sup> Current advancements in radiographic techniques and a deeper realization of IL have increased the possibility of imaging and clinical characterization of the abdominal cystic lesion. However, a range of other abdominal lesions including cystic teratomas, enteric cysts, pancreatic psuedocyst, and alimentary tract duplication may masquerade as lymphangioma.<sup>[5]</sup> In this study, we investigated the evidence of clinic characteristics and imaging features that can contribute to the clinic impression. In addition, we detailed the reasons for misdiagnosis and the therapeutic method of cystic lymphangioma originating in the abdomen treated in our institution, in addition to investigating postoperative recurrences and processing modes.

Acce	Access this article online				
Quick Response Code:	Website: www.cmj.org				
	<b>DOI:</b> 10.4103/0366-6999.169061				

## METHODS

The medical records of 21 patients who were admitted for intra-abdominal lymphangioma between April, 2003 and July, 2013 in our institution were retrospectively analyzed. In our series, IL was commonly located in the mesentery and retroperitoneum areas. The relationship between age, sex, symptoms, physical signs, and location of lymphangioma of each patient were summarized. All patients had an ultrasound (US), abdominal computed tomography (CT) scan and/or magnetic resonance imaging (MRI). For these three examination methods, the number, location, size of cysts, and whether the cysts were unilocular or multilocular, as

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Received: 16-06-2015 Edited by: Peng Lyu How to cite this article: Li Q, Ji D, Tu KS, Dou CW, Yao YM. Clinical Analysis of Intraperitoneal Lymphangioma. Chin Med J 2015; 128:3043-9. well as radiological manifestations of IL, were collected. The correlation between surgical procedure and postoperative complications was investigated. Furthermore, histopathological examinations were intraoperatively performed for all cases. Follow-up information was obtained through clinical interviews, and recurrences were discussed in detail. We stated that the protocol for the research project had been approved by a suitably constituted Ethics Committee of the institution and that it conforms to the provisions of the *Declaration of Helsinki*.

#### **Statistical analysis**

Statistical Package for the Social Sciences (International Business Machines Corporation, New York, NY, USA) version 18.0 was used for descriptive analysis. Continuous variables are presented as the mean  $\pm$  standard deviation (SD). Categorical variables are presented as the number and proportion. We only did the statistical description without statistical analysis because of the few cases.

## RESULTS

Twenty-one cases consisted of retroperitoneal, mesenteric, pancreatic and adrenal cysts, and a cyst in the hepatorenal fossa was also reported [Table 1]. The number of males and females was 10 and 11, respectively, with a sex proportion of 1.1. The mean age was 40 (range 4–80) years. There were five children, with an age range of 4–15 years, and 16 adults (27–80 years) in our

Table 1: Summa	ary of	the	21	patients	with
intraperitoneal	lymph	angi	oma	a	

Variables	Values
Gender (n)	
Male	10
Female	11
Age (years), mean (range)	40 (4-80)
Symptoms ( <i>n</i> )	
Yes	14
No	7
Location ( <i>n</i> )	
Mesentery	7
Retroperitoneum	10
Others	4
Cyst types ( <i>n</i> )	
Unilocular	16
Multilocular	5
Mass size (cm), mean $\pm$ SD	$7.30\pm3.28$
Diagnosis (n)	
Right	19
Wrong	2
Therapy ( <i>n</i> )	
Laparotomy	19
Laparoscopy	1
No	1
Follow-up (years), mean $\pm$ SD	$5.75\pm2.83$
Recurrence ( <i>n</i> )	
Yes	2
No	18

series [Table 2]. The incidence of each age group was shown in Table 3. The analysis based on sex revealed that there was no gender predilection.

Clinically, symptoms happened in 14 of the 21 (67%, 14/21) cases in our series. The relationship between locations and symptoms of the 21 patients with intraperitoneal lymphangioma was summarized in Table 4. The main symptom was intermittent dull abdominal pain, which could be aggravated by the erect position. Fatigue was also found in eight patients (38%, 8/21) (the location of the mass of these eight patients included mesentery [n=3], retroperitoneal [n=3], hepatorenal fossa [n = 1], and pancreas [n = 1]). Three patients (14%, 3/21) complained of persistent abdominal pain, sometimes accompanied by nausea and vomiting when the pain become serious (the locations were, respectively, mesentery [n = 1] and retroperitoneal [n = 2]). Seven patients (33%, 7/21) showed no discomfort, and in four of these cases, either the patient or a family member could touch the lump without assistance (the lumps of those seven patients were located in mesentery [n = 2], retroperitoneal [n = 4], or adrenal gland [n = 1]). The participants had histories of pain varying in duration from 4 days to 3 years. Intestinal obstruction symptoms (abdominal pain and distension, without exhaust) indicated the existence of an abdominal lump in one patient (5%, 1/21) whose diagnosis was mesenteric lymphangioma. One participants with large retroperitoneal cyst complained of diarrhea that persisted for 2 months. A man with acute appendicitis showed right lower quadrant pain in the abdomen with nausea and vomiting, and we found a pancreatic mass in the process of the preoperative imaging examination. The physical examination revealed an abdominal mass in 16 patients (76%, 16/21), and of these, eight patients (50%, 8/16) could feel pain when the location of the IL was pressed. Although a significant percentage of patients had clinical manifestation, there were no positive findings in the physical examination.

Three of the 5 (4–15 years) children (60%, 3/5) complained of the sensation of abdominal pain of varying degrees, and a palpable mass could be touched in the other two kids, who were completely asymptomatic. In these five children, four were mesenteric, and one was retroperitoneal. In the 16 adults (27–80 years), 10 cases (63%, 10/16) described a feeling of discomfort, which included two cases of mesenteric lymphangioma, six cases of retroperitoneal lymphangioma, one case of pancreatic lymphangioma, and one case located in the hepatorenal fossa. The lumps of four patients were found upon routine examination, and no symptoms were present; the lumps were found in the adrenals (n = 1) and retroperitoneum (n=3), respectively. In addition, a man was admitted upon consequence discovering a palpable mass himself, resulting in a diagnosis of renal lymphangioma.

The blood chemistry for all patients was unremarkable. The abdominal US and CT scan were carried out in the 21 intraabdominal cystic lesions that could be the culprit of symptoms and/or signs [Figure 1]. Only one person was checked by MRI, which better clarified the nature of the mass [Figure 2]. Radiological studies revealed a unilocular

Cases No.	Sex, age (years)	Complaint	Duration of symptoms	Physical examination	Diagnosis	Location	Size (cm)
1	Male, 44	Intermittent dull pain	2 years	Palpable mass and deep tenderness	US, CT	Retroperitoneum	$7 \times 6 \times 5$
2	Female, 47	Intermittent dull pain	1 year	Palpable mass	US, MRI	Hepatorenal fossa	$15\times 12\times 8$
3	Female, 46	Feelingless	-	No palpable abnormality	US, CT	Adrenals	$7 \times 7 \times 6$
4	Female, 43	Intermittent dull pain	3 months	No palpable abnormality	US, CT	Retroperitoneum	$6 \times 6 \times 5$
5	Male, 40	Palpable mass	_	Palpable mass	US, CT	Mesentery	$8\times9\times6$
6	Male, 34	Intermittent dull pain aggravated by fatigue	1 month	No palpable abnormality	US, CT	Retroperitoneum	$3 \times 4 \times 4$
7	Male, 40	Persistent pain aggravated by fatigue and erect position	40 days	Palpable mass and deep tenderness	US, CT	Retroperitoneum	$6 \times 6 \times 7$
8	Female, 52	Intermittent dull pain	2 years	Palpable mass and mild tenderness	US, CT	Pancreas	$8 \times 6 \times 4$
9	Female, 67	Persistent pain, nausea and vomiting	6 months	Palpable mass	US, CT	Retroperitoneum	$5 \times 5 \times 3$
10	Male, 54	Intermittent dull pain	1 year	Palpable mass	US, CT	Mesentery	$6\times6\times5$
11	Female, 60	Feelingless	-	No palpable abnormality	US, CT	Pancreas	$10\times 6\times 4$
12	Female, 80	Fever, hypogastralgia and nausea (the cyst superinduced appendicitis)	-	Palpable mass and abdominal pain	US, CT	Retroperitoneum	$8 \times 7 \times 5$
13	Male, 65	Feelingless	-	No palpable abnormality	US, CT	Retroperitoneum	$5\times 6\times 4$
14	Female, 27	Abdominal pain, distension and ceasing exhaust air	2 days	Palpable mass, obvious tenderness and gurgling	US, CT	Mesentery	$10 \times 6 \times 8$
15	Male, 57	Diarrhea	20 days	No palpable abnormality	US, CT	Retroperitoneum	$6 \times 6 \times 4$
16	Female, 49	Palpable mass	-	Palpable mass	US, CT	Retroperitoneum	$4\times 4\times 5$
17	Female, 6	Intermittent dull pain	20 days	Palpable mass	US, CT	Mesentery	$12\times6\times7$
18	Female, 4	Increasing abdominal mass	-	Palpable mass and mild tenderness	US, CT	Mesentery	$8 \times 9 \times 6$
19	Male, 5	Intermittent pain and vomiting	3 days	Obvious tenderness	US, CT	Mesentery	$6 \times 4 \times 3$
20	Male, 15	Persistent pain and abdominal distension aggravated by erect position	4 days	Palpable mass and tenderness	US, CT	Mesentery	$20 \times 20 \times 15$
21	Male, 10	Palpable mass	-	Palpable mass	US, CT	Retroperitoneum	$15\times 10\times 8$
Total	Female:Male = $1.1^*$	-	-	-	-	-	$7.30\pm3.28^{\ddagger}$

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\*The number of male and female was 10 and 11, respectively, with a sex proportion of 1.1; <sup>†</sup>Mean age was 40 (range 4–80) years; <sup>‡</sup>The size of cysts range varied from 3.67 cm to 18.33 cm with an average size of  $7.30 \pm 3.28$  cm. US: Abdominal ultrasound; CT: Abdominal computed tomography scan; MRI: Magnetic resonance imaging; IL: Intraperitoneal lymphangioma. "–" represents that these patients do not have any symptoms.

patients with intraperitoneal lymphangioma								
Age group	Mesentery ( <i>n</i> )	Retroperitoneum (n)	Others (n)	Total ( <i>n</i> , %)				
1-18 years	4	1	0	5 (24)				
Male	2	1	0	3				
Female	2	0	0	2				
19-30 years	1	0	0	1 (5)				
Male	0	0	0	0				
Female	1	0	0	1				
31-50 years	1	5	2	8 (38)				
Male	1	4	0	5				
Female	0	1	2	3				
51-80 years	1	5	1	7 (33)				
Male	1	2	0	3				
Female	0	3	1	4				
Total ( <i>n</i> , %)	7 (33)	11 (52)	3 (14)	21				

Table 3: Distribution of age and location of the 21

Total (n, %)7 (33)11 (52)3 (14)21cyst in 16 patients, and five multilocular masses were defined

cyst in 16 patients, and five multilocular masses were defined by the septae dividing the cystic spaces. The locations of the cysts are summarized in Table 5.



**Figure 1:** Abdominal computed tomography scan (a) and computed tomography contrast enhancement (b) show an intra-abdominal cystic mass. Computed tomography clearly showed a cystic mass with homogeneous internal density and no contrast enhancement after intravenous injection. Retroperitoneal cyst was indicated by red arrow.

In our cases, even in a patient with remote hemorrhage, calcifications were not described. Nineteen cases (90%, 19/21) were accurately diagnosed as lymphangioma prior to surgery based on clinic features and imaging examinations. It is worthwhile to note that two patients were diagnosed other than lymphangioma. As Table 3 illustrated, two patients showed nonspecific symptoms and signs. For example, a woman was diagnosed as having cystadenoma

of the pancreas due to evidence of an enhanced echo on the back wall of the cyst under US and mild enhancement of the wall of the cyst, as finding on abdominal CT. In addition, abundant signaling of blood flow was noted on the abdominal US and a CT revealed that the cyst had an uneven density giving the clinic impression of sole angioma in one boy.

The size of the cysts varied from 3.67 to 18.33 cm, with an average size of  $7.30 \pm 3.28$  cm. The most common site of these cysts was the retroperitoneum (n = 10, 48%, 10/21) with a medial scale of 6.6 cm, followed by the mesentery (n = 7, 33%, 7/21) with a mean size of 9.1 cm. The pancreas (n = 2, 9%, 2/21, 6 cm), adrenals (n = 1, 5%,1/21, 7 cm), and hepatorenal fossa (n = 1, 5%, 1/21, 8 cm) were less frequent locations. Rapid cell smear diagnosis during the operation was implemented for all surgical patients. Histopathological findings were similar in all specimens: Dilated lymphatic vessels, lymph, lymphocytes, and flattened epithelial cells of the cyst inside wall [Table 6].

 Table 4: Symptoms and location of the 21 patients with intraperitoneal lymphangioma

Location	Mesentery ( <i>n</i> )	Retroperitoneum (n)	Others (n)	Total ( <i>n</i> , %)
Symptoms	5	7	2	14 (67)
Feelingless	2	3	2	7 (33)
Total	7	10	4	21

Table 5: Summary of cyst types and location of the 21 patients with intraperitoneal lymphangioma, n (%)

Location	Unilocular ( $n = 16$ )	Multilocular ( $n = 5$ )
Retroperitoneum	9 (56)	2 (40)
Mesentery	5 (31)	2 (40)
Adrenals	1 ( 6)	-
Pancreas	1 (6)	_
Hepatorenal fossa	-	1 (20)

Surgical treatment was performed on 20 patients, while one a 57-year-old man refused the surgery. The primary surgical approach was total excision of the masses by laparotomy performed in 16 patients. In the remaining three patients with mesenteric lymphangioma, the cyst and a small portion of the intestine were removed to excise the mass completely. It is worth mentioning that one cyst, including its base, was treated laparoscopically [Figure 3]. Two patients (10%, 2/21) were in an emergent condition that needed to immediate intervention, a young woman suffered from small bowel obstruction secondary to a large mesenteric mass, and the other presented with acute appendicitis that was complicated by lymphangioma. There were two cysts that had evidence of remote hemorrhage, since the hydatid fluid manifested as brown fluid. It is worth mentioning that we extracted "milky" liquid from a cyst of an elderly woman. In the rest of the 17 patients, the hydatid fluid was clear and flavescent. All cases had an uneventful postoperative course. There were no postoperative mortalities. In all, 17 (85%, 17/20) of the 20 patients were followed up with an average of 5.8 years (2–10 years). Of these, all agreed to be a telephone follow-up. Recurrence of IL at the original site occurred in two patients (10%, 2/20) whose cysts adhered to the surrounding tissues at various degrees. During the first resection, we separated the mass from the surrounding tissues as completely as possible without damaging the normal structures. Therapy after the recurrence included excision of the mass together with the accretive tissues in the two individuals, as shown in Table 7.

## DISCUSSION

#### Location and histological type

IL is a lymphatic malformation, accounting for <5% of all lymphangioma cases.<sup>[6,7]</sup> Some scholars treat this as a hematoma<sup>[8]</sup> with many dilated lymphatic channels demonstrating multiple cystic spaces which is widely regarded as a developmental abnormality.<sup>[9,10]</sup> The most

lable 6: Information of misclagnosis								
Cases	Sex, age (years)	Symptoms signs	US	CT	Initial diagnosis			
1	Female, 52	Intermittent dull pain, palpable mass, and mild tenderness	Enhanced echo of back wall and incomplete capsule	Mild enhancement of the cyst	Cystadenoma of pancreas			
2	Male, 5	Intermittent pain, vomiting, and obvious tenderness	Honeycombing and abundant signal of blood flow	Inside uneven density	Angioma			

CT: Computed tomography; US: Ultrasound.

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Figure 2: Magnetic resonance imaging showing the cystic lesion in retroperitoneum. Coronal images (a and b), sagittal image (c). Magnetic resonance imaging showed the mass as hyperintense on T2-weighted sequences, suggesting fluid content, with regular margins, thin walls, and internal septa. Red arrow referred to be a large cyst in retroperitoneum.

popular sites of IL include the mesentery and retroperitoneum areas for children, but IL may also be acquired later in life after an exogenous insult such as radiation or surgery.<sup>[11-14]</sup> Some existing research states that mesenteric lymphangioma occurs more frequently than retroperitoneal,<sup>[15]</sup> although, in our study, retroperitoneal lymphangioma (48%) had a higher prevalence than did mesenteric lymphangioma (33%). Other possible abdominal sites for the tumor include the liver,<sup>[16]</sup> spleen.<sup>[17]</sup> kidney.<sup>[18]</sup> ligamentum hepatoduodenale.<sup>[6]</sup> gall bladder,<sup>[19]</sup> the falciform ligament,<sup>[20]</sup> and the omentum.<sup>[15]</sup> The true incidence of IL is obscure, and there appears to be no true on sex predilection.<sup>[5]</sup> The findings of our study were consistent with these previous results. Goh et al. also reported that IL has a female preponderance in adult because of endogenous estrogens,<sup>[2]</sup> while some reports demonstrated a male predominance.<sup>[3,21]</sup> Of the three histological types of the lymphangioma described,<sup>[22]</sup> all masses were determined



Figure 3: Resection was performed by laparoscopy. Laparoscopic mass (a and b). Polycyclic edge and internal hydatid fluid was observed during the surgery. Red arrow referred to be a cyst with clear liquid.

Table 7: Management and recurrence of the 20\* II natients who received surgery

to be cysts in our research, although intraperitoneal cavernous masses have been reported in the literatures.<sup>[23]</sup> Nevertheless, the capillary type was found to be absent in the abdomen.

#### Symptoms

In the ordinary course of events, symptoms are stimulated by the primary mass (52%), while in a few circumstances, they are triggered by complications (5%). For instance, in our study, acute abdominal pain and ceasing exhaust air with nausea and vomiting was the result of intestinal obstruction secondary to mesenteric lymphangioma. It is of interest that two patients with intracystic remote hemorrhage did not complain of discomfort. Previous literatures, however, have reported that acute hemorrhage invariably produces acute abdominal diseases.<sup>[24,25]</sup> The mean diameter of the mesenteric cysts  $(8.57 \pm 4.54 \text{ cm})$  was lager compared to the retroperitoneal masses  $(5.83 \pm 2.05 \text{ cm})$ , and consequently, the rate of symptoms in mesentery cysts was 71% and the rate was 42% in retroperitoneum masses. Furthermore, complications such as intestinal obstruction, torsion, and inflammation, are more likely to occur in large lymphangioma.<sup>[26]</sup> The research also states that indisposition induced by masses has something to do with the size which was confirmed in our research.[15,22,27,28]

Our study further considered that the location, compression of adjacent tissues, and distribution of vessels and nerves of the cysts might be the primary cause of symptoms, which were also linked to one's pain threshold. The mechanism of pain might

Cases No.	Location	Surgery	Pathology	Hydatid fluid	Follow-up (years)	Recurrence				
1	Retroperitoneum	Complete resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	6	No				
2	Hepatorenal fossa	Complete resection	Multilocular cyst, flattened endothelial cells, ecstatic lymph vessel	Clear and flavescent liquid	6	No				
3	Adrenal	Complete resection	Unilocular cyst, no lining cells	Clear and flavescent liquid	4	No				
4	Retroperitoneum	Complete resection	Multilocular cyst, flattened endothelial cells	Clear and flavescent liquid	4	No				
5	Mesentery	Complete resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	4	No				
6	Retroperitoneum	Complete resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	2	No				
7	Retroperitoneum	Complete resection by laparoscopy	Multilocular cyst, flattened endothelial cells	Clear and flavescent liquid	3	No				
8	Pancreas	Complete resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	8	No				
9	Retroperitoneum	Complete resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	6	No				
10	Mesentery	Complete resection	Multilocular cyst, flattened endothelial cells	Clear and flavescent liquid	6	No				
11	Retroperitoneum	Complete resection	Unilocular cyst, no lining cells	"Milk" liquid	4	No				
12	Pancreas	Complete resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	2	No				
13	Retroperitoneum	Incomplete resection	Unilocular cyst, flattened endothelial cells	Brown liquid in cyst	2	Yes				
14	Mesentery	Complete resection, mesentery, ileum resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	11	No				
15	Retroperitoneum	Complete resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	7	No				
16	Mesentery	Complete resection, small intestine resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	5	No				
17	Mesentery	Complete resection	Unilocular cyst, flattened endothelial cells	Brown liquid in cyst	10	No				
18	Mesentery	Complete resection, small intestine resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	10	No				
19	Mesentery	Complete resection	Multilocular cyst, flattened endothelial cells	Clear and flavescent liquid	5	No				
20	Retroperitoneum	Incomplete resection	Unilocular cyst, flattened endothelial cells	Clear and flavescent liquid	10	Yes				
The me	ean years of follow	The mean years of follow-up is $5.75 \pm 2.83$ . *A 57-year-old man refused to receive surgery. IL: Intraperitoneal lymphangioma.								

Chinese Medical Journal | November 20, 2015 | Volume 128 | Issue 22

relate to nerve traction on the cyst wall and compression of the adjacent nerve, vessel or organ of the gradually increased mass, which again leads to nonspecific gastrointestinal reactions. These masses remained asymptomatic until reach the threshold described above. Goh *et al.* found that younger patients, including children, were more likely to have obvious and more acute symptoms,<sup>[2,27,29]</sup> and our study substantiates this observation [Table 2]. Symptoms were absent in 33% cases, which was higher than what was noted in the previous research.<sup>[28]</sup>

#### Imaging examination and diagnosis

In a typical US image we may observe fluid in the sonolucent area with the use of enhancement effects, and the boundary of the cysts and internal separate structures are well visible. CT clearly showed the relationship between cystic and adjacent tissues, while typically, contrast administration fails to enhance the imaging of the cyst walls,<sup>[30]</sup> as illustrated in Figure 1. Unfortunately, occasional enhanced capsule walls or abundant flow signals might cause confusion in the diagnosis of lymphangioma. Awareness of this unusual occurrence can lead to a correct diagnosis. In the case of adequate realization of lymphangioma in the abdomen, US combined with CT will allow doctors to make the correct diagnostic decision.

In our study, the accuracy rate of preoperative diagnosis was 90%, which contradicts the literature, which states that preoperative diagnosis is difficult.<sup>[2,24]</sup> In addition, CT is more valuable in the diagnostic process than the other available diagnostic tools.<sup>[30]</sup> MRI is infrequently used for the diagnosis of lymphangioma in the literatures,<sup>[31]</sup> but it enables better preoperative evaluation of intra-abdominal cysts. Clinical, pathologic diagnosis is the gold standard. Pathologically, these lesions may be unilocular or multilocular and contain fluid, which may range from a clear, straw-colored liquid to brown liquid, possibly indicating remote hemorrhage. Generally, the occurrence of these lesions is single, multiple lesions are reported to be a rare event.

#### **Therapy and recurrence**

Surgical resection should be administered immediately after the establishment of a diagnosis of lymphangioma,<sup>[32,33]</sup> since the masses gradually increase in size.<sup>[34,35]</sup> As the masses increase, there is a growing incidence of complications that will appear over time, such as infection, hemorrhage,<sup>[3]</sup> intestinal obstruction (our case), and tumor growth which may prevent complete removal of the cysts, leading to the increased possibility of recurrence and/or loss of adjacent structures.<sup>[32,33]</sup> As described above, three patients lost part of their intestine, and two cases experienced recurrence due to incomplete excision. It should be noted that cystic lymphangioma may give rise to Hodgkin lymphoma.<sup>[36]</sup>

Some surgeons worry about the infiltrative nature of some lesions and the difficulty in achieving complete resection,<sup>[3,11,23]</sup> however, observation of an unambiguous wall is practical, both in radiological studies and during the surgery, and it was feasible to perform complete excision

in 90% of the patients with a recurrence rate of 10% and hardly any morbidity. Incomplete resection was likely responsible for the two cases of recurrences in our study. Hence, it is necessary to excise segmental normal adjacent tissues that appeared to be continuous with the cyst,<sup>[28]</sup> a recommendation that does contradict the recommendation in some previous research.<sup>[37]</sup> Placement of a drainage tube after surgery is essential for preventing chylous ascites. In recent years, laparoscopic excision was given more attention as a potential therapy for intra-abdominal lymphangioma with increasing use and popularity of laparoscopy.<sup>[38]</sup> From our own perspective, laparoscopy is a preferable method if it can accomplish a total resection.

#### **Financial support and sponsorship**

Nil.

#### **Conflicts of interest**

There are no conflicts of interest.

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