Clinical Analysis of Intraperitoneal Lymphangioma

Qing Li, Dong Ji, Kang-Sheng Tu, Chang-Wei Dou, Ying-Min Yao
Department of Hepatobiliary Surgery, The First Affiliated Hospital of Medical College, Xi’an Jiaotong University, Xi’an, Shaanxi 710061, China

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%. Lesions often take place in mesentery, retroperitoneum. Most previously reported ILs are asymptomatic and are often incidentally found through imaging investigation or during surgery for other unrelated causes. 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 pseudocyst, and alimentary tract duplication may masquerade as lymphangioma. 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.

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 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
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 ± 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 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 participant 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

| Variables | Values          |
|-----------|----------------|
| Gender    |                |
| Male      | 10             |
| Female    | 11             |
| Age (years), mean (range) | 40 (4–80) |
| Symptoms  |                |
| Yes       | 14             |
| No        | 7              |
| Location  |                |
| Mesentery | 7              |
| Retroperitoneum | 10        |
| Others    | 4              |
| Cyst types |            |
| Unilocular| 16             |
| Multilocular | 5             |
| Mass size (cm), mean ± SD | 7.30 ± 3.28 |
| Diagnosis |                |
| Right     | 19             |
| Wrong     | 2              |
| Therapy   |                |
| Laparotomy| 19             |
| Laparoscopy| 1             |
| No        | 1              |
| Follow-up (years), mean ± SD | 5.75 ± 2.83 |
| Recurrence|                |
| Yes       | 2              |
| No        | 18             |
Table 2: Clinical characteristics of the 21 intraperitoneal lymphangioma patients

| 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 × 6 × 5 |
| 2         | Female, 47       | Intermittent dull pain | 1 year | Palpable mass | US, MRI | Hepatorenal fossa | 15 × 12 × 8 |
| 3         | Female, 46       | Feelingless | – | No palpable abnormality | US, CT | Adrenals | 7 × 6 × 7 |
| 4         | Female, 43       | Intermittent dull pain | 3 months | No palpable abnormality | US, CT | Retroperitoneum | 6 × 6 × 5 |
| 5         | Male, 40         | Palpable mass | – | Palpable mass | US, CT | Mesentery | 8 × 9 × 6 |
| 6         | Male, 34         | Intermittent dull pain aggravated by fatigue | 1 month | No palpable abnormality | US, CT | Retroperitoneum | 3 × 4 × 4 |
| 7         | Male, 40         | Persistent pain aggravated by fatigue and erect position | 40 days | Palpable mass and deep tenderness | US, CT | Retroperitoneum | 6 × 6 × 7 |
| 8         | Female, 52       | Intermittent dull pain | 2 years | Palpable mass and mild tenderness | US, CT | Pancreas | 8 × 6 × 4 |
| 9         | Female, 67       | Persistent pain, nausea and vomiting | 6 months | Palpable mass | US, CT | Retroperitoneum | 5 × 5 × 3 |
| 10        | Male, 54         | Intermittent dull pain | 1 year | Palpable mass | US, CT | Mesentery | 6 × 6 × 5 |
| 11        | Female, 60       | Feelingless | – | No palpable abnormality | US, CT | Pancreas | 10 × 6 × 4 |
| 12        | Female, 80       | Fever, hypogastalgia and nausea (the cyst superinduced appendicitis) | – | Palpable mass and abdominal pain | US, CT | Retroperitoneum | 8 × 7 × 5 |
| 13        | Male, 65         | Feelingless | – | No palpable abnormality | US, CT | Retroperitoneum | 5 × 6 × 4 |
| 14        | Female, 27       | Abdominal pain, distension and ceasing exhaust air | 2 days | Palpable mass, obvious tenderness and gurgling | US, CT | Mesentery | 10 × 6 × 8 |
| 15        | Male, 57         | Diarrhea | 20 days | No palpable abnormality | US, CT | Retroperitoneum | 6 × 6 × 4 |
| 16        | Female, 49       | Palpable mass | – | Palpable mass | US, CT | Retroperitoneum | 4 × 4 × 5 |
| 17        | Female, 6        | Intermittent dull pain | 20 days | Palpable mass | US, CT | Mesentery | 12 × 6 × 7 |
| 18        | Female, 4        | Increasing abdominal mass | – | Palpable mass and mild tenderness | US, CT | Mesentery | 8 × 9 × 6 |
| 19        | Male, 5          | Intermittent pain and vomiting | 3 days | Obvious tenderness | US, CT | Mesentery | 6 × 4 × 3 |
| 20        | Male, 15         | Persistent pain and abdominal distension aggravated by erect position | 4 days | Palpable mass and tenderness | US, CT | Mesentery | 20 × 20 × 15 |
| 21        | Male, 10         | Palpable mass | – | Palpable mass | US, CT | Retroperitoneum | 15 × 10 × 8 |
| Total     | Female:Male = 1.1* – | – | – | – | – | – | 7.30 ± 3.28‡ |

*The number of male and female was 10 and 11, respectively, with a sex proportion of 1.1; †Mean age was 40 (range 4–80) years; ‡The size of cysts range varied from 3.67 cm to 18.33 cm with an average size of 7.30 ± 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.

Table 3: Distribution of age and location of the 21 patients with intraperitoneal lymphangioma

| Age group   | Mesentery (n) | Retroperitoneum (n) | Others (n) | Total (n, %) |
|-------------|---------------|---------------------|------------|-------------|
| 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 (n, %) 7 (33) 11 (52) 3 (14) 21

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 ± 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 (n) | Retroperitoneum (n) | Others (n) | Total (n, %) |
|----------------|--------------|---------------------|------------|--------------|
| 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)              |

Table 6: Information of misdiagnosis

| 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.

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.

Discussion

Location and histological type

IL is a lymphatic malformation, accounting for <5% of all lymphangioma cases. Some scholars treat this as a hematoma with many dilated lymphatic channels demonstrating multiple cystic spaces which is widely regarded as a developmental abnormality. IL is a lymphatic malformation, accounting for <5% of all lymphangioma cases. Some scholars treat this as a hematoma with many dilated lymphatic channels demonstrating multiple cystic spaces which is widely regarded as a developmental abnormality. 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.
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.\cite{11, 14, 15}

Some existing research states that mesenteric lymphangioma occurs more frequently than retroperitoneal,\cite{13} 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,\cite{16} spleen,\cite{17} kidney,\cite{18} ligamentum hepatoduodenale,\cite{19} gall bladder,\cite{19} the falciform ligament,\cite{20} and the omentum.\cite{15}

The true incidence of IL is obscure, and there appears to be no true on sex predilection.\cite{21} 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,\cite{22} while some reports demonstrated a male predominance.\cite{13, 21} Of the three histological types of the lymphangioma described,\cite{23} all masses were determined to be cysts in our research, although intraperitoneal cavernous masses have been reported in the literatures.\cite{23} 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.\cite{24, 25} The mean diameter of the mesenteric cysts (8.57 ± 4.54 cm) was larger compared to the retroperitoneal masses (5.83 ± 2.05 cm), and consequently, the rate of symptoms in mesenteric 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.\cite{26} The research also states that indisposition induced by masses has something to do with the size which was confirmed in our research.\cite{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

![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.](image)

### Table 7: Management and recurrence of the 20* IL patients who received surgery

| 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         | Mesentry                  | 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        | Mesentry                  | 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        | Mesentry                  | Complete resection, mesentry, ileum resection | Unilocular cyst, flattened endothelial cells | Clear and flavescent liquid | 11                | No         |
| 15        | Retroperitoneum           | Complete resection, small intestine resection | Unilocular cyst, flattened endothelial cells | Clear and flavescent liquid | 7                 | No         |
| 16        | Mesentry                  | Complete resection, small intestine resection | Unilocular cyst, flattened endothelial cells | Clear and flavescent liquid | 5                 | No         |
| 17        | Mesentry                  | Complete resection                           | Unilocular cyst, flattened endothelial cells | Brown liquid in cyst     | 10                | No         |
| 18        | Mesentry                  | Complete resection, small intestine resection | Unilocular cyst, flattened endothelial cells | Clear and flavescent liquid | 10                | No         |
| 19        | Mesentry                  | Complete resection                           | Multilocular cyst, flattened endothelial cells | Clear and flavescent liquid | 5                 | No         |
| 20        | Retroperitoneum           | Complete resection                           | Unilocular cyst, flattened endothelial cells | Clear and flavescent liquid | 10                | Yes        |

The mean years of follow-up is 5.75 ± 2.83. *A 57-year-old man refused to receive surgery. IL: Intraperitoneal lymphangioma.
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,[2,27,29] 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.[28]

**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,[30] 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.[2,24] In addition, CT is more valuable in the diagnostic process than the other available diagnostic tools.[30] MRI is infrequently used for the diagnosis of lymphangioma in the literatures,[31] 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 recent 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, since the masses gradually increase in size.[32,33] As the masses increase, there is a growing incidence of complications that will appear over time, such as infection, hemorrhage,[32] 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.[32,33] 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.[36]

Some surgeons worry about the infiltrative nature of some lesions and the difficulty in achieving complete resection,[31,11,23] 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,[29] a recommendation that does contradict the recommendation in some previous research.[37] 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.[38] 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.

**References**

1. Cutillo DP, Swayne LC, Cucco J, Dougan H. CT and MR imaging in cystic abdominal lymphangiomatosis. J Comput Assist Tomogr 1989;13:534-6.
2. Goh BK, Tan YM, Ong HS, Chui CH, Ooi LL, Chow PK, et al. Intra-abdominal and retroperitoneal lymphangiomas in pediatric and adult patients. World J Surg 2005;29:837-40.
3. de Perrot M, Rostan O, Morel P, Le Coultre C. Abdominal lymphangioma in adults and children. Br J Surg 1998;85:395-7.
4. Reis DG, Rabelo NN, Aratake S. Mesenteric cyst: Abdominal lymphangioma. Arq Bras Cir Dig 2014;27:160-1.
5. Güümüstas OG, Sanal M, Güner O, Tümay V. Retroperitoneal cystic lymphangioma: A diagnostic and surgical challenge. Case Rep Pediatr 2013;2013:292053.
6. Nazarewski L, Patkowski W, Pacho R, Marczewska M, Kruwczyn M. Gall-bladder and hepatoduodenal ligament lymphangioma – Case report and literature review. Pol Przegl Chir 2013;85:39-43.
7. Singh S, Baboo ML, Pathak IC. Cystic lymphangioma in children: Report of 32 cases including lesions atrrare sites. Surgery 1971;69:947-51.
8. Tung KS, McCormack JJ. Angiomatous lymphoid hamartoma. Report of five cases with a review of the literature. Cancer 1967;20:525-36.
9. Bozkaya S, Ugur D, Karaca I, Ceylan A, Uslu S, Baris E, et al. The treatment of lymphangioma in the buccal mucosa by radiofrequency ablation: A case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006;102:e28-31.
10. Mayer M, Fartab M, Villiger A, Yurtsever H. Cystic lymphangioma of the transverse mesocolon. Chirurg 1994;65:561-3.
11. Alqhtani A, Nguyen LT, Flageole H, Shaw K, Laberge JM. 25 years’ experience with lymphangiomas in children. J Pediatr Surg 1999;34:1164-8.
12. Niwa H, Sumita N, Ishihara K, Hoshino T, Iwase H, Kuwabara Y. A case of retroperitoneal chylous cyst developed after cholecystectomy and cholecdothotomy. Nihon Geka Gakkai Zasshi 1988;89:282-5.
13. Gleason TJ, Yuh WT, Tali ET, Harris KG, Mueller DP. Traumatic cervical cystic lymphangioma in an adult. Ann Otol Rhinol Laryngol 1993;102:564-6.
14. Schmidt M. Intra-abdominal lymphangioma. Kans Med Rev 1992;93:149-50.
15. Walker AR, Putnam TC. Omental, mesenteric, and retroperitoneal cysts: A clinical study of 33 new cases. Ann Surg 1973;178:13-9.
16. Stavropoulos M, Vagianos C, Scopa CD, Dragotis C, Androulakis J. Solitary hepatic lymphangioma. A rare benign tumour: A case report. HPB Surg 1994;8:33-6.
17. Castellón Pavón C, Lanchas Alfonso I, González Núñez MA, Amigo Lozano ML, del Amo Olice E. Splenic and adrenal lymphangiomatosis. Rev Esp Enferm Dig 2003;95:585-8.
18. Choi WJ, Jeong WK, Kim Y, Kim J, Pyo JY, Oh YH. MR imaging of hepatic lymphangioma. Korean J Hepatol 2012;18:101-4.
19. Ohba K, Sugauchi F, Orito E, Suzuki K, Ohno T, Mizoguchi N, et al. Cystic lymphangioma of the gall-bladder: A case report. J Gastroenterol Hepatol 1995;10:693-6.
20. Morgan K, Ricketts RR. Lymphangioma of the falciform ligament – A case report. J Pediatr Surg 2004;39:1276-9.
21. Losanoff JE, Richman BW, El-Sherif A, Rider KD, Jones JW. Mesenteric cystic lymphangioma. J Am Coll Surg 2003;196:598-603.
22. Roisman I, Manny J, Fields S, Shiloni E. Intra-abdominal lymphangioma. Br J Surg 1989;76:485-9.
23. Chung JH, Suh YL, Park IA, Jang JJ, Chi JG, Kim YI, et al. A pathologic study of abdominal lymphangiomas. J Korean Med Sci 1999;14:257-62.
24. Allen JG, Riall TS, Cameron JL, Askin FB, Hruban RH, Campbell KA. Abdominal lymphangiomas in adults. J Gastrointest Surg 2006;10:746-51.
25. Bridda A, Dallagnese L, Frego M. Is laparoscopic cholecystectomy safe for lymphangioma of the gallbladder? A complicated case mimicking subhepatic abscess. Updates Surg 2012;64:73-6.
26. Chung JC, Song OP. Cystic lymphangioma of the jejunal mesentery presenting with acute abdomen in an adult. Can J Surg 2009;52:E286-8.
27. Kurtz RJ, Heimann TM, Holt J, Beck AR. Mesenteric and retroperitoneal cysts. Ann Surg 1986;203:109-12.
28. Makni A, Chebbi F, Fetirich F, Ksantini R, Bedioui H, Jouini M, et al. Surgical management of intra-abdominal cystic lymphangioma. Report of 20 cases. World J Surg 2012;36:1037-43.
29. Bliss DP Jr, Coffin CM, Bower RJ, Stockmann PT, Ternberg JL. Mesenteric cysts in children. Surgery 1994;115:571-7.
30. Yang DM, Jung DH, Kim H, Kang JH, Kim SH, Kim JH, et al. Retropertitoneal cystic masses: CT, clinical, and pathologic findings and literature review. Radiographics 2004;24:1353-65.
31. Choi JY, Kim MJ, Chung JJ, Park SI, Lee JT, Yoo HS, et al. Gallbladder lymphangioma: MR findings. Abdom Imaging 2002;27:54-7.
32. Mackenzie DJ, Shapiro SJ, Gordon LA, Ress R. Laparoscopic excision of a mesenteric cyst. J Laparoendosc Surg 1993;3:295-9.
33. Okumus M, Salman T, Gürler N, Salman N, Abbasoglu L. Mesenteric cyst infected with non-typhoidal salmonella infection. Pediatr Surg Int 2004;20:883-5.
34. O TM, Rickert SM, Diallo AM, Scheuermann-Poley C, Otokiti A, Hong M, et al. Lymphatic malformations of the airway. Otolaryngol Head Neck Surg 2013;149:156-60.
35. Adas G, Karatepe O, Atilio M, Battal M, Bender O, Ozcan D, et al. Diagnostic problems with parasitic and non-parasitic splenic cysts. BMC Surg 2009;9:9.
36. Büyükkapu-Bay S, Corapcioğlu F, Gürkan B, Erçin MC, Anik Y, Karadogan M. Mediastinal Hodgkin lymphoma arising from cystic lymphangioma: Case report in a child. Turk J Pediatr 2012;54:298-300.
37. Casadei R, Minni F, Selva S, Marrano N, Marrano D. Cystic lymphangioma of the pancreas: Anatomoclinical, diagnostic and therapeutic considerations regarding three personal observations and review of the literature. Hepatogastroenterology 2003;50:1681-6.
38. Crema E, Etchebehere RM, Gonzaga MN, Lima RS, Bertulucci PA, da Silva AA. Splenic lymphangioma: A rare benign tumor of the spleen treated by laparoscopic surgery. Arq Bras Cir Dig 2012;25:178-9.