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Case Series

Intra-abdominal cystic lymphangioma in adults: A case series of 32 patients and literature review

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ABSTRACT

Introduction: Cystic lymphangioma (CL) is a benign tumor originating from the lymph vessels. Lymphangiomas in the abdominal cavity are extremely rare, particularly in adults.
This article was designed to study the epidemiological, diagnostic difficulties, and therapeutic principles of intra-abdominal cystic lymphangioma (ACL) in adults.
Material and methods: We conducted a single-center, retrospective study of 32 adult patients with ACL admitted to surgical department "A" in "La Rabta Hospital" in Tunis, from January 1998 through December 2020. The demographic, clinical, biological, radiological characteristics, histopathologic, and therapeutic data were collected, as well as the surgical intervention used and the postoperative immediate and late complications.
Results: Thirty-two adult patients with ACL were recruited, including 20 females and 12 males. The median age at treatment was 47 (range 14–80) years. The most prevalent sites were the retroperitoneum (25%), the mesentery (21.9%), and the paracolic gutters (n = 18. 7%). Twenty patients underwent open surgery (62.5%), whereas 12 cases (37.5%) had laparoscopic surgery. Twenty-eight patients received total cystectomy (87%). Three recurrences were observed during follow-up (9.4%).
Conclusion: The clinical features of CL in adults remain unclear. The diagnosis is only confirmed by histopathological examination after complete surgical resection. The laparoscopic approach is considered safe and feasible.

1. Introduction

Cystic lymphangioma is a benign tumor originating from the lymph vessels. It is usually discovered in childhood. The etiology is unclear [1]. It is considered an incorrect embryological connection of the lymphatics when primary lymphatic cysts fail to converge with the main lymphatic system [2,3]. CL may develop in a variety of anatomic locations. The majority of CL locates in the cervical and axial regions. Abdominal cystic lymphangioma (ACL) is a rare entity, representing less than 5% of all CL cases [4]. The clinical presentation is highly polymorphic. The preoperative diagnosis is facilitated using modern imaging. Early diagnosis of abdominal cystic lymphangiomas is important for the surgical treatment of these tumors [5].

In the present study, we aimed to describe the clinical presentations, the complications, the diagnostic modalities, and the therapeutic possibilities of ACL.

2. Materials and methods

We conducted a single-center, retrospective study of adult patients with ACL admitted to the surgical department "A" in "La Rabta Hospital" in Tunis, from January 1998 to December 2020.

The demographic, clinical, biological, radiological characteristics, histopathologic, and therapeutic data were collected, as well as the

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surgical intervention used, and the postoperative immediate and late complications.

The demographic and clinical findings of each patient were documented including age, sex, functional signs, location of CL, symptoms, and physical examination findings. Available ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI) results were reviewed. Radiological characteristics were documented including the number, localization, and size of cysts.

For all patients, the therapeutic approach, the surgical procedure, and postoperative complications were recorded.

The histopathological examination of the resected specimen was consistent with typical findings of a CL.

Follow-up of these patients was performed regularly by a physical examination and abdominal imaging.

All data were recorded and analyzed using IBM SPSS Statistics for Windows, version 25.0 (IBM Corp., Armonk, NY, USA). Descriptive data were expressed in numbers (percentage) for categorical variables and mean (standard deviation, SD) for continuous variables, as appropriate. All data tests were two-sided and a P-value < 0.05 was considered statistically significant.

The work has been reported in line with the PROCESS criteria [6].

3. Results

Thirty-two patients who underwent surgical removal of intra-abdominal CL were included. The median age at diagnosis was 47 (range 14–80) years. There were 20 women and 12 men with a sex ratio equal to 0.6.

Abdominal pain was the main symptom. It was found in 20 patients (62. 5%). Six patients (18. 8%) were asymptomatic and had CL discovered incidentally. For these six patients, the discovery of CL was made preoperatively during an ultrasound for another pathology (n = 5) or intraoperatively during cholecystectomy for a cholelithiasis (n = 1). Other reported signs were constipation (n = 2) due to compression of adjacent organs, dysuria (n = 1), tenesmus and sub occlusive syndrome (n = 1). Fever was reported in 5 cases due to infected CL.

Each patient underwent a detailed physical examination, which revealed an abdominal mass in 15 patients (46.8%). Nine patients (28. 1%) had a normal medical exam. Diagnostic imaging procedures included ultrasound and CT scan (Fig. 1).

The most common site was retroperitoneum (n = 8; 25%) including 3 cases of CL of the Adrenal gland, the mesentery (n = 7; 21.9%), the paracolic gutters (n = 6; 18. 7%), the posterior cavity of the omentum (n = 2; 6.2%), the ileocecal junction (n = 2; 6.2%), the obturator foramen (n = 2; 6.2%), the gastro-splenic ligament (n = 2; 6.2%), the spleen (n = 1; 3.1%), and the abdominal wall (n = 1; 3.1%).

The CL sizes ranged from 2 to 35 cm with a median size of 10 cm. The demographic and clinical characteristics of these patients are shown in Table 1.

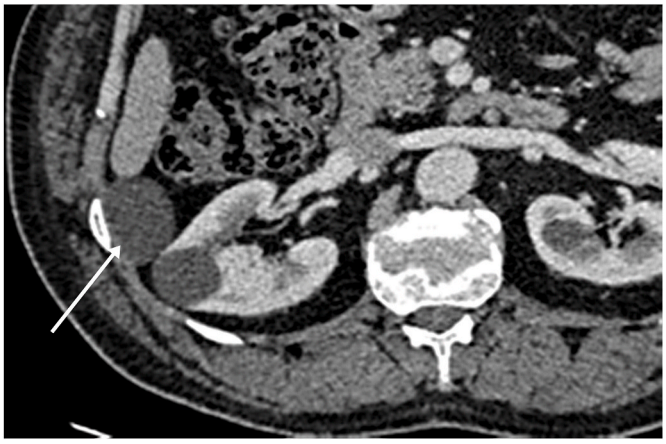

Fig. 1. CT scan showing ACL of the right paracolic gutter (arrow).

The main complications of ACL observed were infection (n = 5; 15,6%), and intracystic active bleeding (n = 1; 3,1%).

Twenty patients underwent open surgery (62.5%) while 12 patients (37.5%) had a laparoscopic surgery (Figs. 2 and 3). No conversion was noted. Surgical treatment consisted of complete resection in 28 patients (87,5%). All our patients were discharged uneventfully.

The histopathologic study matched the diagnosis of CL in all cases: dilated lymphatic vessels, lined with flattened endothelial cells without atypia, and with abundant lymphoid tissue. The diagnosis was confirmed by immunohistochemistry, where the marker D2-40 was positive.

The postoperative follow-up revealed a recurrence of the CL at the operative site in three patients (9.4%), of which only one was reoperated.

Treatments and outcomes for the patients in our cohort are shown in Table 2.

Table 1
Demographic and clinical characteristics of adult CL.

Number	Sex	Age	Symptomatology	Physical sign	Diagnostic means
1	m	37	Abdominal pain	Mass + abdominal distension	US + CT scan
2	f	40	Fortuitous	Absent	US + CTscan
3	f	65	Abdominal pain + occlusion	Mass	US + CTscan
4	m	69	Abdominal pain + constipation	Mass	Us + CTscan
5	m	46	Tenesmus + dysuria	Mass on rectal examination	US
6	m	60	Abdominal pain	Absent	US + CTscan
7	f	70	Abdominal pain	Absent	US + CTscan
8	m	48	Fortuitous	Absent	US + CTscan
9	f	32	Abdominal pain + fever	Abdominal tenderness + distension	US
10	f	46	Fever + mass	Mass	US + CTscan
11	f	24	Mass	Hernia	US
12	f	37	Abdominal pain	Mass	CT scan
13	m	40	Abdominal pain	Mass	Us + CTscan
14	f	48	Mass	Hernia	Us + CTscan
15	f	39	Abdominal pain + fever	Abdominal tenderness	US + CTscan
16	f	80	Mass	Mass + abdominal tenderness	US + CTscan
17	f	14	Fortuitous	Mass	US + CTscan
18	f	61	Mass	Mass	US + CTscan
19	f	50	Fortuitous	Hernia	Peroperative
20	m	75	Abdominal pain	Abdominal tenderness	CT scan
21	m	65	Abdominal pain	Fortuitous	Us + CTscan
22	m	75	Abdominal pain	Abdominal tenderness	CT scan
23	f	61	Abdominal pain	Fortuitous	US + CTscan
24	f	52	Abdominal pain	Fortuitous	CT scan
25	f	61	Fever + mass	Hernia	CT scan
26	m	68	Fortuitous	Mass	US + CTscan
27	m	53	Abdominal pain	Fortuitous	US + CTscan
28	f	47	Abdominal pain	Fortuitous	US + CTscan
29	m	25	Abdominal pain + fever	Abdominal tenderness	US + CTscan
30	f	39	Abdominal pain + mass	Hernia	US + CTscan
31	f	22	Abdominal pain + mass + constipation	Mass	US + CTscan
32	f	45	Abdominal pain	Absent	Intraoperatively

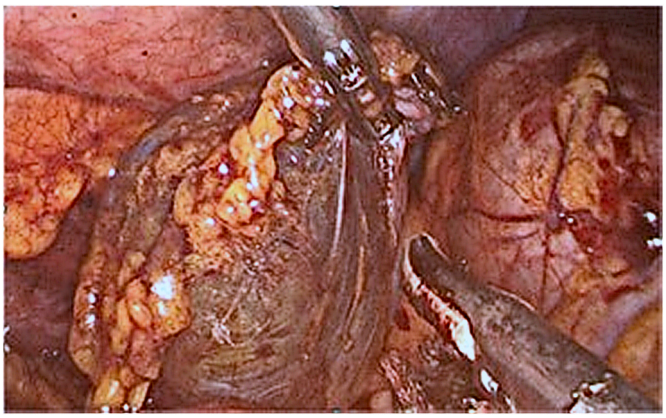

Fig. 2. Laparoscopic view of ACL of the right paracolic gutter.

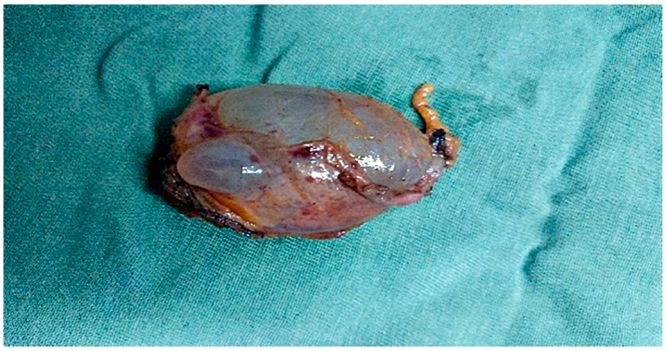

Fig. 3. ACL after total cystectomy.

4. Discussion

To our knowledge, there have only been three cohorts of adults with ACL published in the English literature [5,7,8]. This is the largest adult cohort of patients with ACL described in the literature to date. In our study, the most prevalent sites were the retroperitoneum (25%), the mesentery (21.9%), and the paracolic gutters (n = 18. 7%). Laparoscopy was performed in 37,5% of the patients. By comparing our results with other cohorts, similarities and differences between these cohorts could be drawn (Table 3).

Cystic lymphangioma (CL) is a rare malformative benign tumor of the lymphatic vessels. The etiology of mesenteric lymphangioma is considered to be congenital, with abnormal embryonic development of the lymphatic system causing sequestration of lymphatic tissue [9,10]. Weeda VB et al. suggested they are due to inflammation, trauma, or degeneration [2].

Cystic lymphangiomas is a well-circumscribed cystic lesion with or without endothelial lining, stroma characterized by a meshwork of collagen and fibrous tissue, and a wall containing focal aggregates of lymphoid tissue [11]. More than 80% of lymphangiomas are diagnosed during the first year of life [12–14]. They are rarely observed in adult patients. Men and women are affected similarly in adulthood, whereas our results show a female predominance with a sex ratio equal to 0.6.

Cystic lymphangiomas are preferentially located in the head and the neck (75%), and axilla (20%). They exceptionally occur in the abdomen with less than 10% [15].

Abdominal cystic lymphangiomas occur most commonly in the mesentery, followed by the greater omentum, the mesocolon, and the retroperitoneum [16,17]. In our study, the retroperitoneum was the most prevalent location. Rare cases of ACL in the pancreas, and the stomach have been reported [18,19].

Clinically, most of the cases of cystic lymphangiomas are asymptomatic and are detected incidentally. The clinical manifestations of abdominal cystic lymphangioma are highly polymorphic depending on size and location. In the symptomatic cases, the clinical presentation includes; nausea, vomiting, weight loss, abdominal distension, and acute abdominal pain. In our series, clinical manifestations are variable among patients, with no characteristic signs and symptoms. Fever can be present in patients having infected ACL, as it was the case in our series.

Table 2
Pathological characteristics and surgical management of patients.

Number	Seat	Size	Incision	Gesture	Relapse (years)
1	Mesenteric	25	Laparotomy	Partial cystectomy	0
2	Adrenal	8	laparotomy	Partial cystectomy	1(6)
3	Mesenteric	7	Laparotomy	Total cystectomy	0
4	Mesenteric + mesocolon	12	Laparotomy	Total cystectomy	0
5	Mesocolon	13	Laparotomy	Total cystectomy	0
6	BCO + GS ligament	15	Laparoscopy	Total cystectomy	0
7	GS ligament	8	Laparotomy	Total cystectomy	0
8	Peri renal fat	8	Laparotomy	Total cystectomy	0
9	BCO + spleen	20	Laparoscopy	Partial cystectomy	0
10	Mesenteric	25	Laparotomy	Total cystectomy	1(7)
11	Abdominal Wall	4	Laparotomy	Total cystectomy	0
12	Paracolic gutter	11	Laparoscopy	Total cystectomy	0
13	Adrenal	15	Laparoscopy	Total cystectomy	0
14	Paracolic gutter	2	Laparoscopy	Total cystectomy	0
15	Paracolic gutter	2	Laparotomy	Total cystectomy	0
16	Ileocecal junction	12	Laparotomy	Total cystectomy	0
17	Mesenteric	3	Laparoscopy	Total cystectomy	0
18	Obturator foramen	4	Laparotomy	Total cystectomy	0
19	Retroperit	10	Laparotomy	Partial cystectomy	0
20	Mesenteric	20	Laparotomy	Total cystectomy	0
21	Paracolic gutter	3	Laparoscopy	Total cystectomy	0
22	Mesenteric	10	Laparotomy	Total cystectomy	0
23	Paracolic gutter	20	Laparoscopy	Total cystectomy	0
24	Retroperit	5	Laparoscopy	Total cystectomy	0
25	Obturator foramen	6	Laparotomy	Total cystectomy	0
26	Retroperit	6	Laparoscopy	Total cystectomy	0
27	Retroperit	5	Laparoscopy	Total cystectomy	0
28	Paracolic gutter	5	Laparoscopy	Total cystectomy	0
29	Ileocecal junction	10	Laparotomy	Total cystectomy	0
30	Abdominal wall	10	Laparotomy	Total cystectomy	1(2)
31	Adrenal	30	Laparoscopy	Total cystectomy	0
32	Retroperit	7	Laparoscopy	Partial cystectomy	0

Table 3
Comparison of Tunisian adult ACL cohort with other published cohorts.

Cohorts	Tunisian	French	Chinese
Patients (n)	32	9	12
Male (%)	37.5	10	58
Age at treatment (years)	49	36	39
Locations			
Retroperitoneum (%)	26	66	50
Omentum (%)	6.4	11	17
Mesentery (%)	22	22	17
Spleen (%)	6.4	0	8
Symptoms			
Asymptomatic (%)	19	22	50
Abdominal pain (%)	62	88	33
Physical signs			
Mass touched on abdomen (%)	47	55	15
Imaging Methods			
Ultrasound (%)	84	100	67
CT (%)	84	77	83
MRI (%)	0	22	33
Surgery			
Open surgery (%)	62.5	0	50
Laparoscopic surgery (%)	37.5	100	33
Complete excision (%)	87	100	80
Partial excision (%)	13	0	20
No relapse (%)	90	100	80

Preoperative diagnosis of intra-abdominal lymphangioma is difficult. Ultrasonography, Computed tomography, and Magnetic resonance imaging studies are useful for the diagnosis and surgical planning of abdominal cyst lymphangioma by determining its nature, its location, and its relation to surrounding structures. Among the diagnostic imaging methods, ultrasound is regarded as the first-line examination modality and suitable for disease screening. A thin, clear boundary with strong echo, hypoechoic fluid in the center, and no blood flow signal in color Doppler flow imaging are all typical features of CL. Noticeably, when the cyst is complicated by internal bleeding, it might also have echogenic content [8].

A typical CL under CT is usually a low-density cyst. The shell of the cyst is normally glossy and regularly shaped, while the homogeneous content never takes the contrast. MRI permits to clarify better the nature of the components of the cysts. CL presents as a low signal mass in the T2-weighted image sequence and a high signal in the T1-weighted image sequence [20]. We don't consider that exam necessary. It can be performed in presence of a diagnosis doubt.

The differential diagnoses are lymphoma, digestive duplication, ovarian cyst, hydatid cysts which is endemic in our country, mucinous cystadenomas, and mesenteric cysts [5,21]. But The final proof of the diagnosis of cystic lymphangioma is made by histopathological examination of the specimen [22].

Several modalities of treatments have been proposed.

Asymptomatic patients should be followed by repeated imaging [7,8]. Some lesions may regress spontaneously [8]. The symptomatic lesions can be treated by surgery or percutaneous interventions. The benign nature of the lesion warrants a conservative approach, such as percutaneous aspiration and sclerotherapy. But this strategy is controversial because of the high relapse rate that reach 100% in some reports [7,8,23,24]. In our series, we performed a complete surgical excision of the mass in 87% of the cases which is the treatment of choice for cystic lymphangioma. Complete excision should be secure without causing injuries to the adjacent organs. It should be attempted because of the high recurrence rate after a partial resection [5,25]. Both laparotomy and laparoscopic surgery were reported to treat ACL with acceptable outcomes [7,26–28]. Knowing the advantages of minimally invasive surgery we recommend that the laparoscopic approach should be indicated for most CL. The size of CL does not matter because the cystic fluid can be aspirated, thus decreasing the cystic volume, and creating better working space for the surgeon.

Our study represents the biggest series in the literature describing ACL, their locations, the complications, and the therapeutic modalities. This cohort enriches our knowledge and deepens our understanding of this rare disease.

This study has some limitations that should be pointed out: This is a retrospective study and thus, the evidence level is limited. There is potential for missed recurrences if patients presented to another health structure.

Further prospective studies with larger sample can aid to understand better this pathology and to assess clear therapeutic guidelines.

In summary, CL is a rare benign tumor resulting from a failure in the development of the lymphatic system affecting mainly children. Abdominal localization is rare. The clinical presentation is various. Despite performant imaging technologies, preoperative diagnosis is challenging. Whenever possible, laparoscopic resection should be the treatment of choice. To prevent a recurrence, complete excision is the best option.

Ethical approval

Not required.

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Author contribution

Houcine Maghrebi, Chaima Yakoubi, Hazem Beji, and Feryel Letaief did the conception and design of the work, the data collection, the data analysis and interpretation, and the writing of the manuscript. Sadok Megdich, Seif Boukriba, and Wassim Frikha participated in the writing of the manuscript. Amin Makni, Mouna Ayadi, and Montasser Kacem did the critical revision of the article and the final approval of the version to be published.

Availability of data and materials

All relevant data and materials are provided with in manuscript.

Provenance and peer review

Not commissioned, externally peer reviewed.

Declaration of competing interest

No conflicts of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.104460.

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