Peertechz

CLINICAL GROUP

ARCHIVES OF
Clinical Gastroenterology
OPEN ACCESS JOURNAL

ISSN: 2455-2283 DOI: https://dx.doi.org/10.17352/acg

Case Series

Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review

Avani Saxena[1], Pramath Kakodkar[2]*, Dan Zhang[2], Alysa Poulin[2], Nooshin Shekari[3], Selliah C Kanthan[4] and Rani Kanthan[2]

[1]School of Medicine, University of Saskatchewan, Canada

[2]Pathology and Laboratory Medicine, University of Saskatchewan, Canada

[3]Anatomy, Physiology and Pharmacology, University of Saskatchewan, Canada

[4]Division of Surgery, University of Saskatchewan, Canada

Received: 11 November, 2024
Accepted: 28 November, 2024
Published: 29 November, 2024

***Corresponding author:** Pramath Kakodkar, MD, Pathology and Laboratory Medicine, University of Saskatchewan, Royal University Hospital, 103 Hospital Dr, Saskatoon, SK S7N 0W8, Canada,
E-mail: Pramath.Kakodkar@usask.ca

ORCiD: https://orcid.org/0000-0002-5288-1247

Keywords: Intra-abdominal cystic lymphangioma; Adult mesenteric cysts; Lymphoendothelial cysts; Fibromuscular stroma; Lymphoid aggregates; Cystectomy

https://www.clinsurggroup.us

Check for updates

Abstract

Background: Multi-cystic Lymphangioma (MCL) is a rare intra-abdominal entity in adults. The diagnosis of a core biopsy specimen is challenging due to its rarity in general surgical pathology service. Invasive surgical management can be avoided with accurate core biopsy diagnosis.

Methods: We present six cases of adult intra-abdominal MCL and highlight their clinical, radiological, and pathological characteristics. Demographic, clinical, radiological, and histopathological parameters were collated for analysis.

Results: Six patients (out of 958,574 cases) were identified (prevalence: 0.6/100,000 cases). Previous abdominal surgery (67%, *n* = 4) and abdominal pain with palpable abdominal mass (67%, *n* = 4) at presentation were notable in this cohort. Abdominal CT showed cystic mass with septations, punctate calcification, and fatty halos raising the differential diagnosis of mesenteric cyst (*n* = 2), duplication cyst (*n* = 2), or disseminated metastases (*n* = 2). MCL was distributed across the jejunal mesentery (*n* = 3) and was managed with exploratory laparotomy and resection (*n* = 4). Characteristic histopathological features include multiple variably sized cystic spaces lined by attenuated flattened epithelium, cysts filled with proteinaceous fluid, and interspersed by stroma with several dense lymphoid aggregates. The lymphatic endothelium showed positivity for D2-40 and CD31. Post-operative follow-up at 50.4 ± 49.2 months did not show any clinical or radiological recurrence.

Conclusion: Adult intra-abdominal MCL are rare and radiologically indistinguishable from other intra-abdominal lesions. Diagnostic uncertainty on core biopsy evaluation in our series required invasive surgical exploration. The recognition of the histological triad of lymphoendothelial cysts, smooth-muscle or fibrous stroma, and associated lymphoid aggregates are diagnostic for MCL. Cystectomy alone is curative (144 months without recurrent).

Introduction

Lymphangiomas are designated as lymphatic malformations due to their morphological appearance of haphazard dilatations of lymphatic channels [1]. These lymphangiomas can present in isolation as either single cysts (either with one large cavity or with multiple septae causing a multicystic appearance) or as multiple congruent cysts. Rarely, these lymphangiomas can present as lymphangiomatosis when there are multifocal cystic masses distributed across a broad anatomical region. Lymphangiomas are predominantly seen in the cervical or axillary regions (95%) and the remaining 5% are found in the abdomen (mesentery, retroperitoneum, abdominal organs) and thoracic (lungs and mediastinum) compartments [2,3].

Intra-abdominal multi-cystic lymphangiomas (MCL) are rare and represent less than 5% of all lymphangiomas [4,5]. These are seen in both [6] adults (incidence = 1:175,000) [7-9]

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

and pediatric (incidence = 1:20,000) [7,10] cohorts. Congenital-type pediatric MCL cases are often diagnosed early in life and are linked to developmental errors resulting in lymphatic malformation. Contrastingly, adult MCL cases are extremely rare and suspected to be associated with trauma, previous intra-abdominal surgery, inflammation, or radiation exposure rather than solely congenital. Both the adult and pediatric MCL are considered benign entities, and malignant transformation has not been reported in the scientific literature to date. The rarity of the adult MCL in the general surgical pathology service makes them diagnostically challenging, especially on a core biopsy. Clinically, these often present with abdominal distension [11,12], abdominal pain [13,14], large sized lesions, and most patients do not have co-morbidities. The larger size at presentation can be attributed to the insidious growth pattern of MCL combined with the potential volumetric capacitance within the abdominal cavity.

Radiologically, adult MCLs are seen as anechoic cysts by ultrasonography and as cystic masses with multiple septae by Computer Tomography (CT) [15]. However, pre-operative diagnosis is made in only a minority of patients [16]. These radiological findings are nonspecific and hence it cannot definitively distinguish MCL from the vast differential diagnosis for intra-abdominal cyst (Table 1). Therefore, the definitive diagnosis hinges on the identification of histopathologic features such as dilated spaces lined by lymphoendothelium showing immunohistochemical positivity for D2-40, CD31, and CD34 [17-19]. Laparotomy and segmental enterectomy is the most commonly employed surgical strategy [20] compared to isolated cystectomy [21]. Either strategy results in adequate gross total resection without recurrence [6,16,22] but the invasive surgical option can result in complications such as short gut syndrome [23] Figure 1.

The study aims to report the clinical, radiological, and pathologic features of six cases of adult intra-abdominal MCL and describe their integrated diagnostic insights through a comprehensive literature review. An additional objective is to highlight this rare entity for clinicians and surgical pathologists to enhance and increase the index of suspicion for an accurate diagnosis on core biopsy evaluation.

Case series

An index case of MCL was identified in 2024 which led to a retrospective search for intra-abdominal mesenteric lymphangioma in our laboratory information system since 2000 [24 years]. A total of 958,574 cases were searched which yielded 6 adult patients with MCL. The prevalence of MCL at our institution was estimated at 0.6 per 100,000 cases. Table 1 summarizes the patient demographics, clinical presentation, radiological findings, operative approach, histopathologic features, and clinical follow-up in these 6 patients with MCL. In this cohort, the median age at diagnosis was 58.8 ± 14.6 years (F: M=1:1) and the predominant clinical presentation was abdominal pain with palpable abdominal mass (67%, n = 4). Figure 2 (A-F) illustrates the key radiological image in each of the six cases of MCL. Abdominal CT imaging showed a cystic lesion with thin septation (100%, n = 6) with punctate calcification (50%, n = 3) and fatty halos (33.3%, n = 2). Radiologic differential diagnosis included mesenteric cyst, duplication cyst, lymphoma, disseminated peritoneal carcinomatosis, and advanced mesenteric panniculitis. MCLs were predominantly located in the jejunal mesentery (50%, n = 3), and were managed with exploratory laparotomy (66.7%, n = 4) with cystectomy (66.7%, n = 4).

All 6 cases showed similar histologic findings. Figure 3A shows the masses being composed of variable-sized dilated multicystic spaces [*] which are lined by flattened endothelium. The intervening stroma contains multiple lymphoid aggregates [#] (Figure 3B) and collagenous fibers intermixed with smooth muscle fibers [+] (Figure 3C). There are histologic changes of cyst rupture as seen in Figure 3D by

Table 1: Summary of clinical, radiological, and histopathological parameters in each of the six cases of MCL at our institution.

	Age (Sex)	Case	Radiology (CT or US)	Radiology differential	Operative Procedure	Gross Size (cm)	D2-40	CD31	SMA	Desmin	Follow-up (m)
1	42 (F)	Back pain and palpable mass	Cystic, fatty halos. Scattered calcification foci.	Advanced mesenteric panniculitis, lymphoma, or diffuse metastases	Laparoscopic (cystectomy)	5	+	+	+	+	Stable (6)
2	61 (M)	Abdominal pain and palpable mass	Cystic lesion anterior to infrarenal abdominal aorta	Mesenteric cyst, or duplication cyst	Exploratory laparotomy (cystectomy)	10.5	+	+	+	+	Stable (48)
3	50 (F)	Back and abdominal pain, and palpable mass	Multiseptated cystic mass, layered debris, Calcified	Duplication cyst, Mesenteric cyst of lymphatic or mesothelial origin	Exploratory laparotomy (cystectomy)	NA	+	+	+	+	Stable (144)
4	51 (F)	Abdominal pain, and palpable mass	Complex mesenteric cystic mass, multiseptated with fluid level	peritoneal/ ovarian metastases, or echinococcosis	Exploratory laparotomy (cystectomy)	6	+	+	+	+	Stable (15)
5	88 (M)	Coffee-grounds emesis, and abdominal pain	Perforated viscus. Complex cyst with coarse calcifications	perinephric cyst, or extra-adrenal pheochromocytoma.	Exploratory laparotomy (enterectomy)	0.9	+	+	NA	NA	Death (1)
6	61 (M)	Back pain	Cystic thick-walled lesion with fatty halos and focal coarse calcifications	Retroperitoneal cyst	Interventional radiology drainage	NA	+	NA	NA	NA	Stable (39)

Abbreviations: CT: Computer Tomography; SMA: Smooth Muscle Actin; F: Female; M: Male; m: months.

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

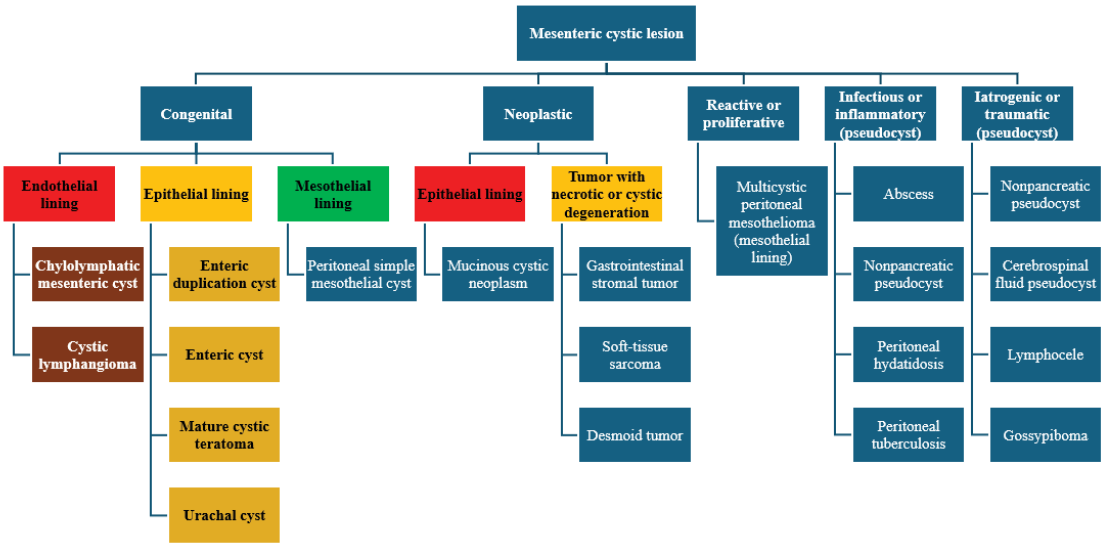

Figure 1: Flowchart summarizing the differential diagnosis of intra-abdominal multicystic lesions.

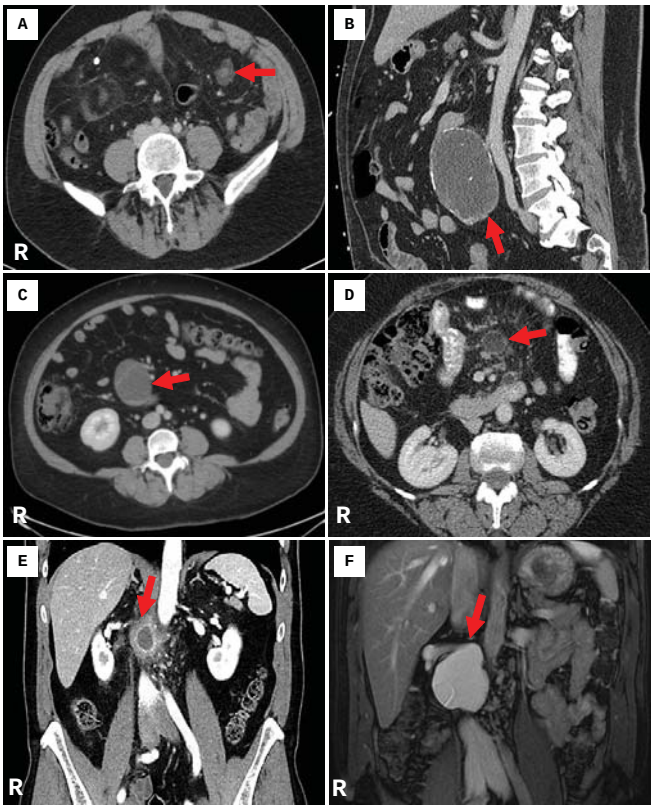

Figure 2: Illustrates the key radiological image in each of the six cases of MCL. A: Case 1- Axial view of contrast-enhanced CT abdomen and pelvis (CTAP) which shows a large hyperdense central mesenteric mass (red arrow) with associated calcification and enlarged nodes. B: Case 2- Sagittal view of contrast-enhanced CTAP which shows a cystic lesion with rim calcification (red arrow) and no cleavage plane with the adjacent duodenum. C: Case 3- Axial view of contrast-enhanced CTAP which shows a minimally complex cystic lesion (red arrow) with a well-circumscribed thin wall, septations, minimal punctate calcifications, and layering debris (proteinaceous or hemorrhagic material). D: Case 4- Axial view of contrast-enhanced CTAP which shows a large mixed-density cystic mass with partial calcification (red arrow). E: Case 5- Coronal view of contrast-enhanced CTAP which shows a peripherally enhancing retroperitoneal cystic mass with coarse calcifications (red arrow). F: Case 6- Coronal view of gadolinium-enhanced MR AP which shows cystic retroperitoneal collection with lobulated margins, and few internal septations.

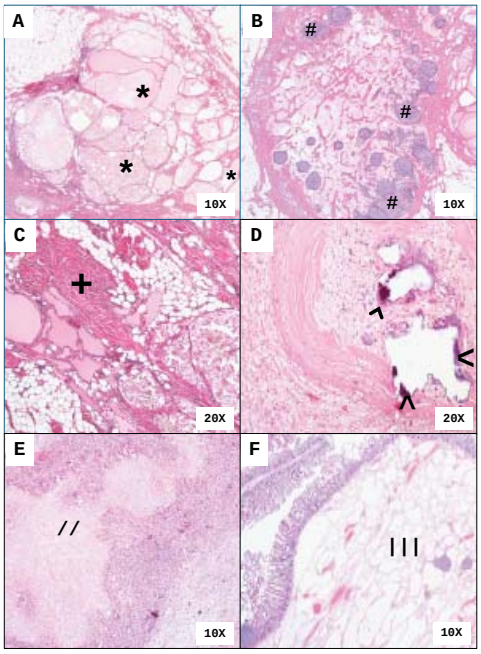

Figure 3: Characteristic microscopic findings of multicystic lymphangioma on hematoxylin and eosin. MCL consists of A: variably sized cyst lined by endothelium [*] and B: lymphoid aggregates [#] with C: intervening stroma made from smooth muscle fibers [+]. MCL can also have D: cyst rupture with calcifications [^] and histiocyte infiltrate. E: Cyst rupture with cholesterol clefts [//] and chronic inflammation. F: MCL [|||] extending into the small intestine submucosa.

the presence of calcification [^], histiocytes and cholesterol clefts [//], and chronic inflammation in Figure 3E. One of the 6 cases from our series showed an MCL involving the submucosa [|||] of the small intestine as seen in Figure 3F.

The immunohistochemistry (IHC) stains for D2-40 and CD31 highlight the cyst lymphatic endothelium lining (Figure 4A and 4B). The intervening stroma shows smooth muscle fibers which stain with SMA and Desmin immunohistochemical antibodies. (Figures 4C,D). There was no expression of Calretinin and

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

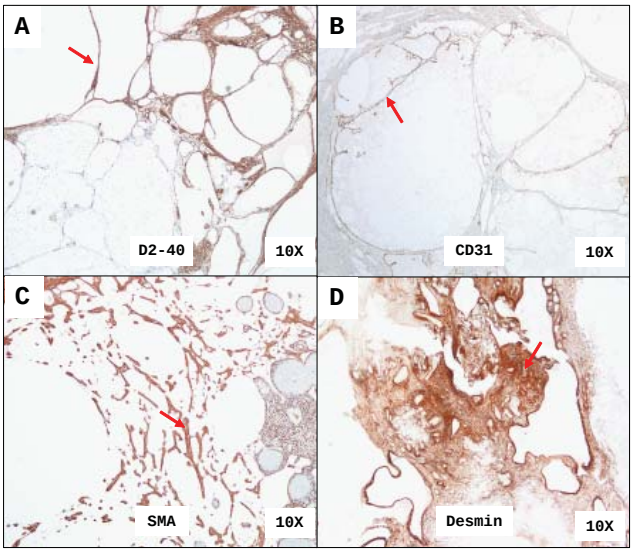

Figure 4: IHC results for MCL. (A) cyst lined by D2-40 (red arrow) and (B) CD31 positive lymphatic endothelium (red arrow). (C) SMA and (D) Desmin highlight smooth muscle fibers (red arrow) within the MCL stroma.

HMB45. The integrated diagnosis based on histology and IHC profile confirms multi-cystic lymphangioma. All 6 patients in the case series remained asymptomatic from MCL recurrence during their follow-up period.

Discussion

Adult intra-abdominal MCL is a rare diagnostic entity in the gastrointestinal clinical and pathology service. A literature review yielded 154 articles in the English language describing intra-abdominal MCL in 118 adult cases [5,8,9,13,16,18-21,24-109] and 133 pediatric cases [5,7,11,12,14,17,23,110-162] indexed in Medline database. It is also notable that the majority were from the United States (n = 61), while the remaining were reported from South Korea (n = 32), France (n = 23), the United Kingdom (n = 20), and Japan (n = 17). We present the second case series on adult intra-abdominal MCL from Canada. Table 2 summarizes the salient clinical, radiological, and pathologic features in adult and pediatric populations. Supplemental Tables S1.1 - S2.2 show the comprehensive clinical, radiological, and pathologic features dataset for adult and pediatric MCL cases respectively.

Our literature review showed that intra-abdominal MCL was identified in both adults (mean: 40.36 years [range: 18-82]) and children (mean: 5.57 years [range: 0-17]). Overall, there was a slight male preponderance; however, in adult MCL cases there was a slight female predilection (M: F ratio = 1:1.2). Contrastingly, the pediatric cohorts showed a male predilection (M: F ratio 1.7:1). Lymphangiomas are considered malformations or hamartomas, and not true neoplasms. This embryonal malformation theory is partially supported by the fact that most cases are diagnosed during early childhood and a large number are diagnosed prenatally [163,164]. Genetic factors are considered to play a role in the pathogenesis of this entity including somatic mutations [165], the role of PIK3CA mutations [166], and in hereditary lesions the role of

Table 2: Summary of the salient clinical, radiological, and pathologic features of Intra-Abdominal Multi cystic Lymphangiomas in adult and pediatric populations in the 154 articles from the literature review restricted to the English Language.

	Adult (n = 118)	Pediatric (n = 133)	p - value adjusted
Clinical Presentation			
Symptomatic at presentation	101 (85.6%)	96 (72.2%)	0.01 (NS)
Abdominal pain	74 (62.7%)	68 (51.1%)	0.06 (NS)
Abdominal distention	19 (16.1%)	24 (18.1%)	0.68 (NS)
Radiologic appearance			
Multiloculated cyst	56 (47.5%)	31 (23.3%)	< 0.0001*
Uniloculated cyst	5 (4.2%)	4 (3.0%)	0.74 (NS)
Multicystic	12 (10.2%)	1 (0.8%)	NA
Surgical Management Options			
Laparotomy segmental enterectomy	50 (42.4%)	65 (48.9%)	0.30 (NS)
Laparotomy cystectomy	60 (51.9%)	40 (30.1%)	< 0.0001*
Laparoscopic segmental enterectomy	1 (0.9%)	1 (0.8%)	NA
Laparoscopic cystectomy	1 (0.9%)	1 (0.8%)	NA
Radiotherapy	1 (0.9%)	0	NA
Cyst drainage and biopsy	0	3 (2.3%)	NA
Cyst size: mean; range	13.7 cm (0.4-50)	12.8 cm (3-26)	0.69 (NS)
Content of the cyst			
Chylous	44 (37.3%)	22 (16.5%)	< 0.0001*
Serous	20 (17%)	18 (13.5%)	0.45 (NS)
Hemorrhagic	10 (8.4%)	9 (6.8%)	0.61 (NS)
Microscopic findings			
Dilated lymphatic channels	112 (94.9%)	130 (97.7%)	0.31 (NS)
Fibro-collagenous stroma with smooth muscle fibers	88 (74.6%)	87 (65.4%)	0.11 (NS)
Lymphoid aggregates	55 (46.6%)	64 (48.1%)	0.81 (NS)
Chronic inflammatory cells	37 (31.4%)	25 (18.8%)	0.02 (NS)
Microcalcifications	6 (5.1%)	5 (3.8%)	0.61 (NS)
Cholesterol clefts	4 (3.4%)	24 (19%)	< 0.0001*
Follow up status			
follow-up (months, range)	14.1 months (1-120)	23 months (0.5-192)	0.41 (NS)
Stable	64 (54.2%)	83 (62.4%)	0.19 (NS)
Death	0	3 (2.3%)	NA
Progression, Recurrence	1 (0.8%)	1 (0.8%)	NA
Short gut syndrome	0	1 (0.8%)	NA

Each row represents a feature analyzed for differences between the two groups. For categorical variables, either a Chi-square test or Fisher's Exact Test was used, depending on the expected cell counts. A Chi-square test was performed if all expected frequencies were ≥ 5; otherwise, Fisher's Exact Test was applied. For continuous variables (cyst size and follow-up), a Mann-Whitney U test was conducted as the dataset was not normally distributed. A Bonferroni correction was applied to account for multiple comparisons across 22 tests. The threshold for statistical significance after adjustment was set at α-adjusted = 0.00227. Features with a p - value < 0.00227 were deemed statistically significant (*), indicating strong evidence for differences between adult and pediatric groups.
Abbreviations: NS: Not Significant; NA: Not Applicable

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

VEGFR-3 [167] encoding genes is implicated. Other possible etiologies include a traumatic origin resulting in bleeding or inflammation in the lymphatic channels, both leading to obstruction and subsequent secondary lymphangiomatous formation [168-170].

All six of our patients were symptomatic, which is in keeping with the overall finding of symptomatic clinical presentation in adults (85.59%) and children (72.18%) MCL in the literature. Abdominal pain is the most common presenting feature, followed by abdominal distension. Abdominal pain in the pediatric cohort is often described as more severe [115] and may be associated with a more acute presentation [7,140,171,172], especially if associated with a rapidly growing mass [140]. Contrastingly, adult patients with MCL present with a chronic pain of more than a few days/weeks duration [171]. Other findings at presentation include nausea and vomiting [30], fever, and features of co-morbidities if present, such as those of anemia [48]. Often a palpable mass is identified, usually related to the location of the lesion [110].

In our case series majority of the adult MCL cases showed multiple septated cysts and calcifications. Abdominal ultrasound and CT are common initial radiologic investigations in these patients. Ultrasonography is very sensitive and relatively specific for the evaluation of abdominal cystic masses, as shown in one study where the sensitivity was 87.9% and specificity 81.8% [173]. The lesions appear as sharply defined cystic or multicystic formations, often with internal septations [2,174,175]. CT findings for MCL demonstrated a predominant homogenous cystic lesion but fluid, blood, or fat may lead to heterogenous imaging findings [176]. Our literature review indicated that the most common finding was intra-abdominal multiloculated cysts (n = 29, 24.58%). Interestingly, multiloculation as a radiological feature was more frequently identified in adults compared to pediatric patients (47.5% *vs.* 23.3%, p - value <0.0001). This higher incidence could be from a combination of developmental progression, later diagnosis, radiologic detection sensitivity in ultrasound *versus* CT, and secondary complications (cyst infection, trauma or hemorrhage). Magnetic resonance imaging (MRI) has been shown to be more helpful because of its high resolution and its ability to delineate cystic and septal structures [22]. However, the usefulness of radiologic investigations for accurate preoperative diagnosis is limited [16] due to the overlap of findings with other cystic lesions in the abdomen [122,177-179] leading to a range of differential radiological diagnoses including disseminated carcinomatosis in adults.

Lymphangiomas are subdivided into three main types: capillary, cavernous, and cystic. The first two are predominantly cutaneous lesions; cystic lymphangiomas are generally found in the abdomen and retroperitoneum. All six of our cases were intra-abdominal MCLs. The literature identifies that most intra-abdominal MCLs are seen in the small bowel mesentery proper (jejunum followed by ileum, or unspecified location of the small intestine) and then in the colonic region (Figure 5). Overall, adult and pediatric MCLs can occasionally present with complications such as mesenteric vessel dilatation (n = 13, 5%) or bowel obstruction (n = 30, 10.7%) due to volvulus, ischemia, malrotation, internal hernia, and/or intussusception.

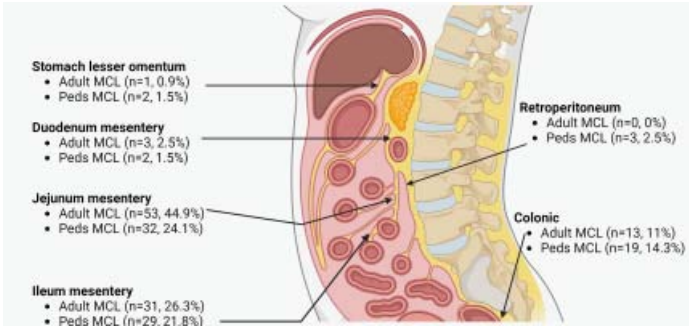

Figure 5: Summary of the anatomic distribution of intra-abdominal multi-cystic lymphangiomas within our literature review (154 articles).

Macroscopically the size of the intra-abdominal MCLs in our case series was approximately 5.6 ± 3.4 cm and were predominantly hemorrhagic (n = 2) in cyst content. Contrastingly, the average cyst size reported in our literature review was 13.7 cm in adults and 12.8 cm in the pediatric cohort. Overall, most cysts have a chylous fluid (n = 32, 26.3%), followed by serous fluid (n = 18, 15%), and hemorrhagic fluid in others (n = 9, 7.5%). Chylous fluid is milky because of the fluid's abundant fat content, while the serous fluid is clear, and straw-colored in appearance. This variability in cyst fluid composition may be multifactorial and related to variable lymph stasis, the number of lymphatic communicating channels, and their fluid protein content [175,180,181]. Interestingly, most intra-abdominal MCLs in the literature were reported to be intact at surgery (n = 174, 98%) with a minority being ruptured (n = 5, 2%). Moreover, the chylous fluid was more frequently identified at grossing in adult MCL compared to pediatric MCL (37.3% *vs.* 16.5%, p - value <0.0001). This higher incidence could be from a multitude of factors. One such possibility is that in adults these lymphangiomas may have more established connections to other lymphatic channels and the thoracic duct resulting in leakage of chyle into these cystic spaces.

Our cases had endothelium-lined variable-size channels, smooth muscle fibers, and lymphoid aggregates in the wall, and some cases especially post-biopsy resected specimens had microcalcifications with inflammatory infiltrates and cholesterol clefts. The endothelium showed immunohistochemical positivity for D2-40 and CD31 and was negative for mesothelial immunohistochemical markers such as calretinin and WT1. Smooth muscle fibers were positive for SMA and Desmin. The histopathological triad for MCLs that were consistently identified in our case series and the literature included dilated lymphatic channels (n = 242, 96.4%), fibro-collagenous stroma with smooth muscle fibers (n = 175, 69.7%), and lymphoid follicles (n = 119, 47.4%). In addition, scattered chronic inflammatory infiltrate (n = 62, 24.7%), microcalcifications (n = 11, 4.4%), and cholesterol clefts (n = 28, 11.1%) have also been reported and some were concerning previous fine needle or core biopsy. Microcalcifications were associated with chylous or hemorrhagic contents and were seen especially in lesions located mostly in the jejunum [34,49].

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

The immunohistochemical profile is essential in diagnosing MCL from its differential diagnosis; lymphoma, adenocarcinoma, and hemangioma if there is secondary bleeding into the abdominal cavity. In our case series, the core biopsy on Case 1 raised the possibility of angiomyolipoma due to the histologic findings of vessels, muscle, and fat. It is important to consider the location of angiomyolipoma and IHC profile to rule out this differential. MCLs usually express markers seen in the lymphatic lining (D2-40, CD31, and CD34) [17-19]. The absence of CD34 [55,70] and factor VIII-related antigen (FVIII-RAG) [64] in the cyst lining has been documented in rare cases but these could be attributed to underlying preanalytical methodological issues. Similarly for other differential diagnoses, specific markers will help with the diagnosis e.g., mesothelioma will have positivity for calretinin, lymphoma for hematolymphoid markers, and adenocarcinoma for epithelial markers. A summary of the IHC findings in the adult and pediatric intra-abdominal MCLs in the literature review is shown in Figure 6.

The extent of surgical resection of intra-abdominal MCLs is dependent on the observation that these tend to involve the surrounding areas and therefore may potentially recur if incompletely excised [174,182]. Common surgical modalities include laparotomy with segmental enterectomy or laparotomy with cystectomy [171,183]. Adjunct surgical procedures after laparotomy are rare, but these depend on the location and extent of the lesion e.g. Whipple's [51], splenectomy [146], and cholecystectomy [100]. Laparoscopic removal is a feasible option in children [184]. Laparoscopic removal of intra-abdominal MCLs has certain advantages over classical laparotomy, including a more precise dissection around the mesenteric vessels, minimal trauma to the abdominal structures, less pain, and a shorter hospital stay. More experience is needed to demonstrate the superiority of this method over conventional open techniques [185]. Interval elective surgery is recommended in patients with the asymptomatic presentation with documented interval regression [28]. Notably, laparoscopic cystectomy was more frequently performed in adult MCL compared to pediatric MCL (51.9% *vs.* 30.1%, p -value < 0.0001). Adult MCL tends to be more localized, allowing for complete cyst removal without bowel resection. Furthermore, segmentectomy has higher risks and functional consequences in adults, making cystectomy preferable. Lastly, pediatric surgical practice tends to have more experience with segmental resections rather than cystectomy.

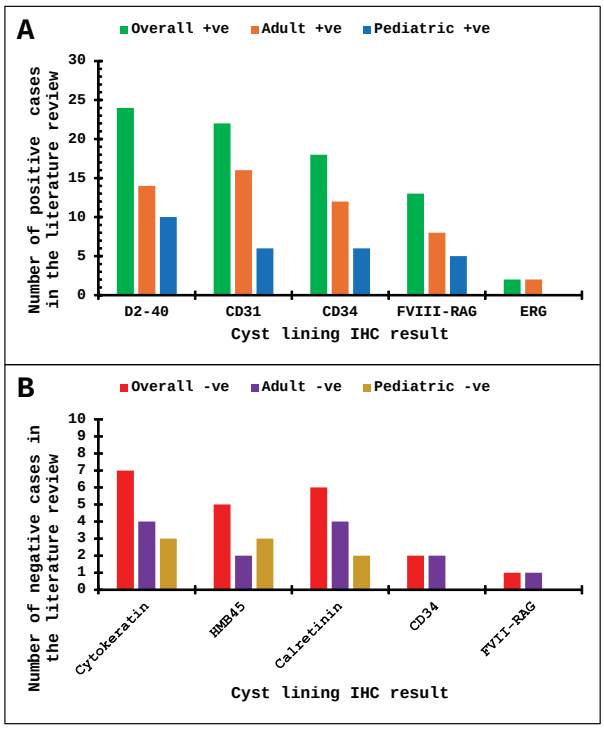

Figure 6: Summary of positive (A) and negative (B) IHC findings for the cyst lining in the overall (adult and pediatric), adult-only, and pediatric-only intra-abdominal MCL cohort within the literature review.

Follow-up was reported in 153 patients (65 adults and 88 pediatric) in the literature; of these, 147 patients were reported as stable (average: 14 months for adults, 23 months for pediatric patients). Most cases do not have recurrences and remain stable [6,16,22,171,186]. Macroscopically complete resection, compared to other procedures, has the lowest rate of recurrence [4]. Patients with limited surgical resection were also noted to be stable in the literature. In one pediatric case series, a high rate of complications, including infections (approximately 6.6%) required additional interventional surgical procedures [182].

Adverse outcomes on follow-up in the literature have been observed only in 6 cases in the literature review. In one adult case, there was a small region of recurrence or possible residual lymphangioma noted on MRI at 6 months post-resection [20]. One pediatric patient had initial progression on ultrasonography at 6 months following incomplete surgical resection due to the risk of short bowel syndrome and proximity of the superior mesenteric artery [17]. It is possible that a combination of the findings of rupture, large size, multicystic nature, fibrous adhesions, and microcalcifications might be related to recurrence and progression [17,20]. The remaining four cases involved short gut syndrome in one child [23] and deaths in three children [23,127,142]. Notably, one adult patient was diagnosed with a large unresectable mass and was treated with the mammalian target of rapamycin (mTOR) inhibitor everolimus (activation of the mTOR pathway was reported at the pathological and transcriptomic levels). This patient eventually had a successful resection of the residual tumor following a major partial response to everolimus [32].

In the literature, the clinical presentation, radiologic findings, cyst size, cyst contents, histologic findings, immunohistochemical features, and follow-up were similar in adults and children. The only notable difference was slight female preponderance in adults and the reverse in children. Given these similarities, the best approach to diagnosis in both age groups is a biopsy and the use of histology and immunohistochemistry to demonstrate lymphatic endothelial lined cysts (D2-40 and CD31) for diagnosis. A minimally invasive surgical procedure appears to be adequate in most cases as the patients are stable on follow-up for both extensive

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

and minimal surgery. A limitation of this literature review is that not all relevant findings such as clinical presentation, radiologic findings, cyst size, detailed gross, and histologic and immunohistochemical descriptions were reported in the published individual case reports and case series.

Conclusion

Adult intra-abdominal MCL is a rare entity that is clinically and radiologically indistinguishable from other intra-abdominal lesions. Due to diagnostic uncertainty on core biopsy evaluation, all cases in our series required invasive surgical exploration. The recognition of the histological triad of lymphoendothelial cysts, smooth-muscle or fibrous stroma, and associated lymphoid aggregates is essential to making a diagnosis of MCL. Our cohort shows that limited surgical intervention such as cystectomy without resection of adjacent organs is curative (144 months without recurrent).

Ethical approval

Standard ethical approval [E-Bio-021] has been collected and preserved by the author(s).

Author contribution

All authors contributed significantly towards this manuscript and have read and approved the final version of the manuscript.

(Supplementary)

References

1. Khunger N. Lymphatic malformations: Current status. J Cutan Aesthet Surg. 2010;3(3):137-8. Available from: https://doi.org/10.4103/0974-2077.74487
2. Lugo-Olivieri CH, Taylor GA. CT differentiation of large abdominal lymphangioma from ascites. Pediatr Radiol. 1993;23(2):129-30. Available from: https://doi.org/10.1007/bf02012405
3. Al-Salem AH. Lymphangiomas in infancy and childhood. Saudi Med J. 2004;25(4):466-9. Available from: https://pubmed.ncbi.nlm.nih.gov/15083217/
4. Alqahtani A, Nguyen LT, Flageole H, Shaw K, Laberge JM. 25 years' experience with lymphangiomas in children. J Pediatr Surg. 1999;34(7):1164-8. Available from: https://doi.org/10.1016/s0022-3468(99)90590-0
5. Farrell WJ, Grube P. Intra-abdominal cystic lymphangiomas. Am J Surg. 1964;108:790-3. Available from: https://doi.org/10.1016/0002-9610(64)90033-9
6. Mede A, Chotai PN, Huh WJ, Tan M. Intra-abdominal cystic lymphangiomas: The Vanderbilt experience. J Surg Res. 2023;285:197-204. Available from: https://doi.org/10.1016/j.jss.2022.12.026
7. de Perrot M, Rostan O, Morel P, Le Coultre C. Abdominal lymphangioma in adults and children. Br J Surg. 1998;85(3):395-7. Available from: https://doi.org/10.1046/j.1365-2168.1998.00628.x
8. Du Y, Zhang JN, Zhu LL, Wang Y, Li WP. Haemolymphangioma of the small bowel mesentery in adults: two case reports and a literature review. BMC Gastroenterol. 2021;21(1):273. Available from: https://doi.org/10.1186/s12876-021-01855-w
9. Reis DG, Rabelo NN, Aratake SJ. Mesenteric cyst: abdominal lymphangioma. Arq Bras Cir Dig. 2014;27(2):160-1. Available from: https://doi.org/10.1590/s0102-67202014000200016
10. Bliss DP Jr, Coffin CM, Bower RJ, Stockmann PT, Ternberg JL. Mesenteric cysts in children. Surgery. 1994;115(5):571-7. Available from: https://pubmed.ncbi.nlm.nih.gov/8178256/
11. Troum S, Solis MM. Mesenteric lymphangioma causes bowel obstruction in a child. South Med J. 1996;89(8):808-9. Available from: https://doi.org/10.1097/00007611-199608000-00011
12. Méndez-Gallart R, Solar-Boga A, Gómez-Tellado M, Somoza-Argibay I. Giant mesenteric cystic lymphangioma in an infant presenting with acute bowel obstruction. Can J Surg. 2009;52(3):E42-3. Available from: https://pubmed.ncbi.nlm.nih.gov/19503641/
13. Losanoff JE, Kjossev KT. Mesenteric cystic lymphangioma: unusual cause of intra-abdominal catastrophe in an adult. Int J Clin Pract. 2005;59(8):986-7. Available from: https://doi.org/10.1111/j.1368-5031.2005.00554.x
14. Poroes F, Petermann D, Andrejevic-Blant S, Labgaa I, Di Mare L. Pediatric cystic lymphangioma of the retroperitoneum: A case report and review of the literature. Medicine (Baltimore). 2020;99(28):e20827. Available from: https://doi.org/10.1097/md.0000000000020827
15. Levy AD, Cantisani V, Markku M. Abdominal lymphangiomas: Imaging features with pathologic correlation. Am J Roentgenol. 2004;182(6):1485-91. Available from: https://doi.org/10.2214/ajr.182.6.1821485
16. Allen JG, Riall TS, Cameron JL, Askin FB, Hruban RH, Campbell KA. Abdominal lymphangiomas in adults. J Gastrointest Surg. 2006;10(5):746-51. Available from: https://doi.org/10.1016/j.gassur.2005.10.015
17. Clement C, Snoekx R, Ceulemans P, Wyn I, Matheï J. An acute presentation of pediatric mesenteric lymphangioma: a case report and literature overview. Acta Chir Belg. 2018;118(5):331-5. Available from: https://doi.org/10.1080/00015458.2017.1379802
18. Mehmedovic Z, Mehmedovic M, Custovic MK, Sadikovic A, Mekic N. A rare case of giant mesenteric cystic lymphangioma of the small bowel in an adult: A case presentation and literature review. Acta Gastroenterol Belg. 2016;79(3):491-3. Available from: https://pubmed.ncbi.nlm.nih.gov/28209109/
19. 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(3):257-62. Available from: https://doi.org/10.3346/jkms.1999.14.3.257
20. Parker DR, Kiely P, Smith R. Complete resection of a massive mesenteric lymphangioma in an adult. BMJ Case Rep. 2020;13(3). Available from: https://doi.org/10.1136/bcr-2019-233714
21. Losanoff JE, Richman BW, El-Sherif A, Rider KD, Jones JW. Mesenteric cystic lymphangioma. J Am Coll Surg. 2003;196(4):598-603. A vailable from: https://doi.org/10.1016/s1072-7515(02)01755-6
22. Su CM, Yu MC, Chen HY, Tseng JH, Jan YY, Chen MF. Single-center results of treatment of retroperitoneal and mesenteric cystic lymphangiomas. Dig Surg. 2007;24(3):181-5. Available from: https://doi.org/10.1159/000102896
23. Chang TS, Ricketts R, Abramowsky CR, Cotter BD, Steelman CK, Husain A, et al. Mesenteric cystic masses: a series of 21 pediatric cases and review of the literature. Fetal Pediatr Pathol. 2011;30(1):40-4. Available from: https://doi.org/10.3109/15513815.2010.505623
24. Sana L, Fehd K, Imen R, Faouzi C. Mesenteric cystic lymphangioma. Surgery. 2021;170(5):e25-e6. Available from: https://doi.org/10.1016/j.surg.2021.04.038
25. Torashima Y, Yamaguchi J, Taniguchi K, Fujioka H, Shimokawa I, Izawa K, et al. Surgery for ileal mesenteric lymphangioma during pregnancy: case report and review of the literature. J Gastrointest Surg. 2004;8(5):616-20. Available from: https://doi.org/10.1016/j.gassur.2003.10.012
26. Talarico F, Iusco D, Negri L, Valieri L. Mesenteric cystic lymphangioma treated with laparoscopic excision: case report and review of the literature. G Chir.

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

2009;30(8-9):362-4.
Available from: https://pubmed.ncbi.nlm.nih.gov/19735616/

27. Protopapas A, Papadopoulos D, Rodolakis A, Markaki S, Antsaklis A. Mesenteric lymphangioma presenting as adnexal torsion: case report and literature review. J Clin Ultrasound. 2005;33(2):90-3.
Available from: https://doi.org/10.1002/jcu.20094

28. Tan DTM, Chok AY, Farah BL, Yan YY, Toh EL. Spontaneous partial regression of a microcystic jejunal mesenteric lymphangioma and a proposed management algorithm. BMJ Case Rep. 2019;12(11).
Available from: https://doi.org/10.1136/bcr-2019-231037

29. Zhang Y, Yang P, Chen M. Cavernous mesenteric lymphangioma presenting as intra-abdominal malignancy. Asian J Surg. 2023;46(11):4855-6.
Available from: https://doi.org/10.1016/j.asjsur.2023.05.135

30. Singh N, Singh R, Maheswari U, Aga P. Primary mesenteric lymphangioma in a young adult with intestinal malrotation and 'counter-clockwise barber pole sign'. BMJ Case Rep. 2013;2013.
Available from: https://doi.org/10.1136/bcr-2013-008994

31. Rajput D, Srikanth K, Gupta A, Kumar A, Edem S, David LE, et al. Large retroperitoneal cystic lymphangioma mimicking mesenteric cyst: a case report. Pan Afr Med J. 2022;42:115.
Available from: https://doi.org/10.11604/pamj.2022.42.115.30777

32. El Zein S, Gruel N, Bonvalot S, Mir O, Watson S. Neoadjuvant Everolimus for Adult Giant Mesenteric Cystic Lymphangioma with mTOR Pathway Activation. Oncologist. 2021;26(7):554-7.
Available from: https://doi.org/10.1002/onco.13775

33. Hureibi K, Sunidar OA. Mesenteric cystic lymphangioma mimicking malignancy. BMJ Case Rep. 2014;2014.
Available from: https://doi.org/10.1136/bcr-2014-203560

34. Bosker RJ, Eddes EH. A mesenteric lymphangioma showing calcification and ossification. Dig Surg. 2004;21(3):182-3.
Available from: https://doi.org/10.1159/000079343

35. Tomsett AL, Addison RE, Hopkins JC, Courtney ED. Small intestinal and mesenteric lymphangioma in an adult: a rare cause of acute abdominal pain. Br J Hosp Med (Lond). 2016;77(10):603.
Available from: https://doi.org/10.12968/hmed.2016.77.10.603

36. Waters EG. Mesenteric lymphangioma with intestinal obstruction complicating uterine carcinoma and fibromyoma. Am J Obstet Gynecol. 1946;52:478-83.
Available from: https://doi.org/10.1016/s0002-9378(15)30265-9

37. Durgakeri P, Penington B. Cystic mesenteric lymphangioma: a case report. Anz J Surg. 2018;88(12):E861-e2.
Available from: https://doi.org/10.1111/ans.13950

38. Wei MY, Chua J, Cheng Y, Grossberg P. Small bowel volvulus in an adult with mesenteric lymphangioma and ascariasis. Anz J Surg. 2018;88(12):E859-e60.
Available from: https://doi.org/10.1111/ans.13953

39. Xu X, Zheng C, He X, Zhao Y, Hong T. Gastrointestinal: Giant mesenteric cystic lymphangioma. J Gastroenterol Hepatol. 2017;32(2):290.
Available from: https://doi.org/10.1111/jgh.13542

40. Chim H, Chuwa E, Chau YP, Chow PK. Gastrointestinal: mesenteric cystic lymphangioma. J Gastroenterol Hepatol. 2006;21(5):916.
Available from: https://doi.org/10.1111/j.1440-1746.2006.04479.x

41. Chin S, Kikuyama S, Hashimoto T, Tomita T, Hasegawa T, Ohno Y. Lymphangioma of the jejunal mesentery in an adult: a case report and a review of the Japanese literature. Keio J Med. 1993;42(1):41-3.
Available from: https://doi.org/10.2302/kjm.42.41

42. Jayasundara J, Perera E, Chandu de Silva MV, Pathirana AA. Lymphangioma of the jejunal mesentery and jejunal polyps presenting as an acute abdomen in a teenager. Ann R Coll Surg Engl. 2017;99(3):e108-e9.
Available from: https://doi.org/10.1308/rcsann.2017.0012

43. Jeong WK, Kim Y, Song SY, Heo JN, Park CK. Cavernous mesenteric lymphangioma (2006: 4b). Eur Radiol. 2006;16(7):1625-8.
Available from: https://doi.org/10.1007/s00330-005-0106-0

44. Liu L, Ke G, Shen WM, Jia CK. Giant cystic lymphangioma in the mesoileum: A case report and literature review. Turk J Gastroenterol. 2020;31(8):603-6.
Available from: https://doi.org/10.5152/tjg.2020.19256

45. Nagano H, Kimura T, Iida A, Togawa T, Goi T, Sato Y. Cystic lymphangioma in the peripheral jejunal mesentery in an adult and excision with laparoscopic-assisted surgery: a case report. World J Surg Oncol. 2019;17(1):170.
Available from: https://doi.org/10.1186/s12957-019-1713-6

46. Paramythiotis D, Bangeas P, Karakatsanis A, Iliadis A, Karayannopoulou G, Michalopoulos A. Ideal treatment strategy for chylous mesenteric cyst: a case report. J Med Case Rep. 2018;12(1):317.
Available from: https://doi.org/10.1186/s13256-018-1716-x

47. Siddique K, Bhandari S, Basu S. Giant mesenteric lymphangioma: a rare cause of a life-threatening complication in an adult. BMJ Case Rep. 2010;2010. Available from: https://doi.org/10.1136/bcr.04.2010.2896

48. Tai PT, Jewell LD. Case report: mesenteric mixed haemangioma and lymphangioma; report of a case with 10-year follow-up after radiation treatment. Br J Radiol. 1995;68(810):657-61.
Available from: https://doi.org/10.1259/0007-1285-68-810-657

49. Thiam O, Faye PM, Niasse A, Seye Y, Gueye ML, Sarr IS, et al. Cystic mesenteric lymphangioma: A case report. Int J Surg Case Rep. 2019;61:318-21. Available from: https://doi.org/10.1016/j.ijscr.2019.07.051

50. Yoo E, Kim MJ, Kim KW, Chung JJ, Kim SH, Choi JY. A case of mesenteric cystic lymphangioma: fat saturation and chemical shift MR imaging. J Magn Reson Imaging. 2006;23(1):77-80.
Available from: https://doi.org/10.1002/jmri.20474

51. Epstein HS, Berman R. Mesenteric and pancreatic lymphangioma presenting as a right adnexal mass. Am J Obstet Gynecol. 1975;121(8):1117-8.
Available from: https://doi.org/10.1016/s0002-9378(16)33602-x

52. Macnab I, Menzies T. Lymphangioma of mesentery. Br J Surg. 1950;37(147):294-8.
Available from: https://doi.org/10.1002/bjs.18003714709

53. Mordi A, Rabii K, Hameed E, Lefrancq T. Intestinal obstruction complicating a mesenteric cystic lymphangioma. J Visc Surg. 2012;149(5):e356-8. Available from: https://doi.org/10.1016/j.jviscsurg.2012.04.001

54. Teng Y, Wang J, Xi Q. Jejunal hemolymphangioma: A case report. Medicine (Baltimore). 2020;99(4):e18863.
Available from: https://doi.org/10.1097/md.0000000000018863

55. Tsuboi M, Noda H, Watanabe F, Abe I, Nokubi M, Rikiyama T. Complete resection of a complicated huge mesenteric lymphangioma guided by mesenteric computed tomography angiography with three-dimensional reconstruction: report of a case. Int Surg. 2015;100(3):574-8.
Available from: https://doi.org/10.9738/intsurg-d-14-00112.1

56. Wall KC, Schmitz R, Carney JM, Blazer III DG. Large mesenteric lymphangioma in an adult patient: an unusual presentation of a rare disease. BMJ Case Rep. 2018;2018.
Available from: https://doi.org/10.1136/bcr-2018-226319

57. Ahn KS, Han HS, Yoon YS, Kim HH, Lee TS, Kang SB, et al. Laparoscopic resection of nonadrenal retroperitoneal tumors. Arch Surg. 2011;146(2):162-7. Available from: https://doi.org/10.1001/archsurg.2010.342

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

58. Albayrak Y, Albayrak F, Arslan S, Calik I. Mesenteric calcified cystic lymphangioma in an adult patient. Turk J Gastroenterol. 2011;22(3):341-3. Available from: https://doi.org/10.4318/tjg.2011.0224

59. Grodek L, Korczyńska-Tartanus B, Bielecki K, Zmora J, Malinowska M, Dmoch-Gajzlerska E. Cystic lymphangioma arising from the small intestine mesentery incidentally found during surgery for a large ovarian tumor - A case report. Wiad Lek. 2022;75(9 pt 1):2170-3. Available from: https://doi.org/10.36740/wlek202209122

60. Jang JH, Lee SL, Ku YM, An CH, Chang ED. Small bowel volvulus induced by mesenteric lymphangioma in an adult: a case report. Korean J Radiol. 2009;10(3):319-22. Available from: https://doi.org/10.3348/kjr.2009.10.3.319

61. Limdi JK, Mehdi S, Sapundzieski M, Manu M, Abbasi AM. Cystic lymphangioma of the mesocolon. J Gastrointest Surg. 2010;14(9):1459-61. Available from: https://doi.org/10.1007/s11605-010-1176-0

62. Miele L, Pierconti F, Forgione A, Vero V, Cammarota G, Molinari F, et al. Cystic lymphangioma of the mesentery and hyposplenism in celiac disease. Eur J Gastroenterol Hepatol. 2007;19(11):1026-30. Available from: https://doi.org/10.1097/meg.0b013e328220ecbd

63. Naganuma H, Ishida H, Komatsuda T, Hakamada M, Sawada T, Satoyoshi R, et al. Sonographic findings in two cases of lymphangioma of the mesocolon in adults. J Clin Ultrasound. 2018;46(1):78-81. Available from: https://doi.org/10.1002/jcu.22488

64. Okamoto D, Ishigami K, Yoshimitsu K, Irie H, Tajima T, Nishie A, et al. Hemorrhagic mesenteric cystic lymphangioma presenting with acute lower abdominal pain: the diagnostic clues on MR imaging. Emerg Radiol. 2009;16(4):327-30. Available from: https://doi.org/10.1007/s10140-008-0747-9

65. Rieker RJ, Quentmeier A, Weiss C, Kretzschmar U, Amann K, Mechtersheimer G, et al. Cystic lymphangioma of the small-bowel mesentery: case report and a review of the literature. Pathol Oncol Res. 2000;6(2):146-8. Available from: https://doi.org/10.1007/bf03032366

66. Springer MR, Rowe PW. Mesenteric lymphangioma. A report of a calcified lymphangioma in the small bowel mesentery. Radiology. 1963;80:954-6. Available from: https://doi.org/10.1148/80.6.954

67. Stein M, Hsu RK, Schneider PD, Ruebner BH, Mina Y. Alcohol ablation of a mesenteric lymphangioma. J Vasc Interv Radiol. 2000;11(2 Pt 1):247-50. Available from: https://doi.org/10.1016/s1051-0443(07)61473-0

68. Târcoveanu E, Moldovanu R, Bradea C, Vlad N, Ciobanu D, Vasilescu A. Laparoscopic treatment of intraabdominal cystic lymphangioma. Chirurgia (Bucur). 2016;111(3):236-41. Available from: https://pubmed.ncbi.nlm.nih.gov/27452935/

69. Vennarecci G, Ceribelli C, Laurenzi A, Moroni E, Ettorre GM. Giant cavernous mesenteric lymphangioma in adult. Updates Surg. 2013;65(4):317-9. Available from: https://doi.org/10.1007/s13304-012-0157-0

70. Wang J, Fisher C, Thway K. Combined mesothelial cyst and lymphangioma of the small bowel: a distinct hybrid intra-abdominal cyst. Int J Surg Pathol. 2014;22(6):547-51. Available from: https://doi.org/10.1177/1066896913509014

71. Blanco Velasco G, Tun Abraham A, Hernández Mondragón O, Blancas Valencia JM. Hemolymphangioma as a cause of overt obscure gastrointestinal bleeding: a case report. Rev Esp Enferm Dig. 2017;109(3):213-4. Available from: https://pubmed.ncbi.nlm.nih.gov/28256143/

72. Botey M, Muñoz-Ramos C, Bonfill J, Roura J, Torres M, Mocanu S, et al. Acute abdomen for lymphangioma of the small bowel mesentery: a case report and review of the literature. Rev Esp Enferm Dig. 2015;107(1):39-40. Available from: https://pubmed.ncbi.nlm.nih.gov/25603331/

73. Clendenning WE, Block JB, Radde IG. Basal cell nevus syndrome. Arch Dermatol. 1964;90:38-53. Available from: https://doi.org/10.1001/archderm.1964.01600010044011

74. Ha TK, Paik SS, Lee KG. Cystic lymphangioma arising from mesocolon. Clin Gastroenterol Hepatol. 2009;7(3):e14-5. Available from: https://doi.org/10.1016/j.cgh.2008.09.013

75. Hornick JL, Fletcher CD. Intraabdominal cystic lymphangiomas obscured by marked superimposed reactive changes: clinicopathological analysis of a series. Hum Pathol. 2005;36(4):426-32. Available from: https://doi.org/10.1016/j.humpath.2005.02.007

76. Hwang SS, Choi HJ, Park SY. Cavernous mesenteric lymphangiomatosis mimicking metastasis in a patient with rectal cancer: a case report. World J Gastroenterol. 2009;15(31):3947-9. Available from: https://doi.org/10.3748/wjg.15.3947

77. Iqbal J, Isaacs P, Sissons M, Bury R, Khurshid M, Akhtar M. A cause for concern? An asymptomatic mesenteric lesion. Cavernous lymphangioma. Gut. 2009;58(9):1184, 225. Available from: https://doi.org/10.1136/gut.2008.166769

78. Lawson JA, Dietrick RB, Russi S. Cystic lymphangioma of the mesentery; review of the literature and report of a case. AMA Arch Surg. 1958;76(1):155-9. Available from: https://doi.org/10.1001/archsurg.1958.01280190157030

79. Miliaras S, Trygonis S, Papandoniou A, Kalamaras S, Trygonis C, Kiskinis D. Mesenteric cyst of the descending colon: report of a case. Acta Chir Belg. 2006;106(6):714-6. Available from: https://doi.org/10.1080/00015458.2006.11679990

80. Oh C, Danese CA, Dreiling DA. Chylous cysts of mesentery. Arch Surg. 1967;94(6):790-3. Available from: https://doi.org/10.1001/archsurg.1967.01330120044009

81. Ryu WS, Kwak JM, Seo UH, Kim SH, Park SS, Kim CS, et al. Laparoscopic treatment of a huge cystic lymphangioma: partial aspiration technique with a spinal needle. J Laparoendosc Adv Surg Tech A. 2008;18(4):603-5. Available from: https://doi.org/10.1089/lap.2007.0145

82. Seki H, Ueda T, Kasuya T, Kotanagi H, Tamura T. Lymphangioma of the jejunum and mesentery presenting with acute abdomen in an adult. J Gastroenterol. 1998;33(1):107-11. Available from: https://doi.org/10.1007/s005350050053

83. Xu X, Liu W, Zheng C. A rare cause of repeated gastrointestinal bleeding. Mesenteric cavernous lymphangioma. Gastroenterology. 2014;146(4):e11-3. Available from: https://doi.org/10.1053/j.gastro.2013.11.040

84. Izzo L, Galati G, Sassayannis PG, Binda B, D'Arielli D, Stasolla A, et al. Mesenteric cystic lymphangioma causing intestinal obstruction in an adult patient. G Chir. 2003;24(10):347-9. Available from: https://pubmed.ncbi.nlm.nih.gov/14722994/

85. Campbell WJ, Irwin ST, Biggart JD. Benign lymphangioma of the jejunal mesentery: an unusual cause of small bowel obstruction. Gut. 1991;32(12):1568. Available from: https://doi.org/10.1136/gut.32.12.1568

86. Chen CW, Hsu SD, Lin CH, Cheng MF, Yu JC. Cystic lymphangioma of the jejunal mesentery in an adult: a case report. World J Gastroenterol. 2005;11(32):5084-6. Available from: https://doi.org/10.3748/wjg.v11.i32.5084

87. Chera R, Gupta AA, Bailey D, Somers GR, Kukreti V, Crump M. Small intestinal B-cell lymphoma in a patient with lymphangiectasia secondary to abdominal lymphangioma. J Clin Oncol. 2008;26(4):675-8. Available from: https://doi.org/10.1200/jco.2007.14.4311

88. Christofi N, Hextall A. Cystic cavernous lymphangioma of the mesentery in a patient with Cowden syndrome. J Obstet Gynaecol. 2007;27(3):329-30. Available from: https://doi.org/10.1080/01443610701269267

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

89. Chung JC, Song OP. Cystic lymphangioma of the jejunal mesentery presenting with acute abdomen in an adult. Can J Surg [Can J Surg]. 2009;52(6):E286-8.
Available from: https://pubmed.ncbi.nlm.nih.gov/20011166/

90. Covarelli P, Arena S, Badolato M, Canonico S, Rondelli F, Luzi G, et al. Mesenteric chylous cysts simulating a pelvic disease: a case report. Chir Ital. 2008;60(2):319-22.
Available from: https://pubmed.ncbi.nlm.nih.gov/18689186/

91. Csak T, Folhoffer A, Horvath A, Halász J, Diczházi C, Schaff Z, et al. Holmes-Adie syndrome, autoimmune hepatitis, and celiac disease: a case report. World J Gastroenterol. 2006;12(9):1485-7.
Available from: https://doi.org/10.3748/wjg.v12.i9.1485

92. Iwabuchi A, Otaka M, Okuyama A, Jin M, Otani S, Itoh S, et al. Disseminated intra-abdominal cystic lymphangiomatosis with severe intestinal bleeding. A case report. J Clin Gastroenterol. 1997;25(1):383-6.
Available from: https://doi.org/10.1097/00004836-199707000-00022

93. Kok KY, Mathew VV, Yapp SK. Lymphangioma of the small-bowel mesentery: unusual cause of intestinal obstruction. J Clin Gastroenterol. 1997;24(3):186-7. Available from: https://doi.org/10.1097/00004836-199704000-00015

94. Majewski M, Lorenc Z, Krawczyk W. Multiannual abdominal pain complicated by obstruction of the gastrointestinal tract in the course of cystic form of mesenteric lymphangioma of the small intestine. Polski Przegląd Chirurgiczny. 2015;87:185-8. Available from: https://pubmed.ncbi.nlm.nih.gov/?term=Lorenc+Z&cauthor_id=26146118

95. Nizami S, Mohiuddin K, Daudi I, Ahmed Z, Memon MA. Cavernous transverse mesocolonic lymphangioma in an adult. Am J Surg. 2007;193(6):740-1.
Available from: https://doi.org/10.1016/j.amjsurg.2006.06.037

96. Norris JR, Stacey M, Rampaul RS, Cheung KL. Jejunal lymphangioma presenting as an ovarian mass. J R Army Med Corps. 2008;154(4):243-4.
Available from: https://doi.org/10.1136/jramc-154-04-07

97. Ratajczak A, Szmeja J, Lukaszuk M, Majewski P. Lymphatic angioma of mesentery of the small intestine: a case report. Pol Przegl Chir. 2011;83(1):48-50.
Available from: https://doi.org/10.2478/v10035-011-0007-7

98. Sadola E. Cystic lymphangioma of the jejunal mesentery in an adult. J Clin Ultrasound. 1987;15(8):542-3.
Available from: https://doi.org/10.1002/jcu.1870150807

99. Shah A, Moftah M, Morrin M, Redmond M, Cahill RA. Single-site laparoscopic excision of mesocolic cystic lymphangioma - a video vignette. Colorectal Dis. 2014;16(7):566. Available from: https://doi.org/10.1111/codi.12589

100. Shikano T, Takeda S, Sakai M, Sugimoto H, Kanazumi N, Nomoto S, et al. Cystic lymphangioma of the gallbladder: report of a case. Surg Today. 2008;38(1):81-4.
Available from: https://doi.org/10.1007/s00595-007-3564-y

101. Shimura H, Ueda J, Ogawa Y, Ichimiya H, Tanaka M. Total excision of mesenteric cysts by laparoscopic surgery: report of two cases. Surg Laparosc Endosc. 1997;7(2):173-6.
Available from: https://pubmed.ncbi.nlm.nih.gov/9109254/

102. Thompson GJ, Culp OS. Perplexing cystic masses near the kidney. J Urol. 1963;89:370-6.
Available from: https://doi.org/10.1016/s0022-5347(17)64560-9

103. Tomizawa Y, Garner K, Sohnen A. Lymphangioma of the small bowel mesentery: a rare intra-abdominal tumor causing anemia. Clin Gastroenterol Hepatol. 2013;11(8):E57.
Available from: https://doi.org/10.1016/j.cgh.2012.11.013

104. Tsukada H, Takaori K, Ishiguro S, Tsuda T, Ota S, Yamamoto T. Giant cystic lymphangioma of the small bowel mesentery: report of a case. Surg Today. 2002;32(8):734-7.
Available from: https://doi.org/10.1007/s005950200138

105. Viar WN, Scott WF Jr, Donald JM. Mesenteric cavernous lymphangiomata: brief review and report of two cases. Ann Surg. 1961;153(1):157-60.
Available from: https://doi.org/10.1097/00000658-196101000-00019

106. Vogel TR, Hammond JS, Schwarz RE. Image of the month--giant multicystic lymphangioma. Arch Surg. 2005;140(4):411-2. Available from: https://doi.org/10.1001/archsurg.140.4.411

107. Zilko PJ, Laurence BH, Sheiner H, Pollard J. Cystic lymphangiomyoma of the colon causing protein-losing enteropathy. Am J Dig Dis. 1975;20(11):1076-80. Available from: https://doi.org/10.1007/bf01071198

108. Banks PA, Rosenberg AE. Case 5-1989 — A 27-Year-Old Greek Woman with Portal-Vein Thrombosis and an Epigastric Mass. N Engl J Med. 1989;320(5):301-10.
Available from: https://doi.org/10.1056/nejm198902023200508

109. Field ES. Cavernous chylangioma of jejunal mesentery. Br Med J. 1971;2(5752):27-8. Available from: https://doi.org/10.1136/bmj.2.5752.27

110. Bishton RL, Sames CP, Wallis HR. Calcified mesenteric lymphangioma. Arch Dis Child. 1958;33(168):176-8. Available from: https://doi.org/10.1136/adc.33.168.176

111. Buccoliero AM, Castiglione F, Maio V, Morelli C, Martin A, Messineo A, et al. Calcified cystic lymphangioma of the mesentery: case report. Fetal Pediatr Pathol. 2009;28(5):209-15.
Available from: https://doi.org/10.3109/15513810903073203

112. Hanganu E, Gavrilescu SL, Trandafirescu MF, Chiforeanu AM, Mihăilă D, Florea ID, et al. A histopathological diagnosis of mesenteric cystic lymphangioma, clinically misdiagnosed as simple mesenteric cyst - case report. Rom J Morphol Embryol. 2017;58(4):1525-30. Available from: https://pubmed.ncbi.nlm.nih.gov/29556652/

113. Hardin WJ, Hardy JD. Mesenteric cysts. Am J Surg. 1970;119(6):640-5.
Available from: https://doi.org/10.1016/0002-9610(70)90232-1

114. Kerkeni Y, Zouaoui A, Thamri F, Boujelbene N, Jouini R. Cystic lymphangioma of the small bowel mesentery in a child. J Pediatr. 2022;244:248-9.
Available from: https://doi.org/10.1016/j.jpeds.2021.12.032

115. Kim SH, Kim HY, Lee C, Min HS, Jung SE. Clinical features of mesenteric lymphatic malformation in children. J Pediatr Surg. 2016;51(4):582-7.
Available from: https://doi.org/10.1016/j.jpedsurg.2015.11.021

116. Ko SF, Ng SH, Shieh CS, Lin JW, Huang CC, Lee TY. Mesenteric cystic lymphangioma with myxoid degeneration: unusual CT and MR manifestations. Pediatr Radiol. 1995;25(7):525-7.
Available from: https://doi.org/10.1007/bf02015784

117. Kurbedin J, Haines L, Levine MC, Dickman E. When fever, leukocytosis, and right lower quadrant pain is not appendicitis. Pediatr Emerg Care. 2017;33(9):e46-e7.
Available from: https://doi.org/10.1097/pec.0000000000001250

118. Kusuma FSP, Poerwadi P. Chylous mesenteric cyst in children - A case report in a 4-year-old boy. Med J Malaysia. 2020;75(Suppl 1):48-50.
Available from: https://pubmed.ncbi.nlm.nih.gov/32471968/

119. Leland HA, Lee JT, Tan JH, Romine LE, Bansal V. Cystic lymphangioma of the lesser curvature of the stomach--case report. J Radiol Case Rep. 2011;5(5):31-7. Available from: https://doi.org/10.3941/jrcr.v5i5.716

120. Li H, Bai YZ. Calcified mesenteric lymphangioma mimicking a teratoma in a 4-year-old child. Pediatr Neonatol. 2016;57(3):256-8. Available from: https://doi.org/10.1016/j.pedneo.2015.07.004

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

121. Meyer T, Stöhr G, Post S, Fayyazi A. Retroperitoneal lymphangioma presenting as a mesenteric cyst. Eur J Radiol. 1995;21(2):143-4. Available from: https://doi.org/10.1016/0720-048x(95)00710-8

122. Molander ML, Mortensson W, Udén R. Omental and mesenteric cysts in children. Acta Paediatr Scand. 1982;71(2):227-9. Available from: https://doi.org/10.1111/j.1651-2227.1982.tb09404.x

123. Morgan K, Ricketts RR. Lymphangioma of the falciform ligament--a case report. J Pediatr Surg. 2004;39(8):1276-9. Available from: https://doi.org/10.1016/j.jpedsurg.2004.04.032

124. Niwa Y, Imai K, Kotani T, Nakano T, Ushida T, Moriyama Y, et al. A pitfall in diagnosing fetal abdominal lymphangioma: a report of two cases. J Clin Ultrasound. 2019;47(8):494-6. Available from: https://doi.org/10.1002/jcu.22756

125. Suthiwartnarueput W, Kiatipunsodsai S, Kwankua A, Chaumrattanakul U. Lymphangioma of the small bowel mesentery: a case report and review of the literature. World J Gastroenterol. 2012;18(43):6328-32. Available from: https://doi.org/10.3748/wjg.v18.i43.6328

126. Takeuchi S, Yamaguchi M, Sakurai M, Nonaka K, Awazu S. A case of mesenteric cyst diagnosed by ultrasound examination and a review of Japanese literatures. Jpn J Surg. 1979;9(4):359-65. Available from: https://doi.org/10.1007/bf02468637

127. Walker-Smith JA, Reye RD, Soutter GB, Kenrick KG. Small intestinal lymphangioma. Arch Dis Child. 1969;44(236):527-32. Available from: https://doi.org/10.1136/adc.44.236.527

128. Weeda VB, Booij KA, Aronson DC. Mesenteric cystic lymphangioma: a congenital and an acquired anomaly? Two cases and a review of the literature. J Pediatr Surg. 2008;43(6):1206-8. Available from: https://doi.org/10.1016/j.jpedsurg.2008.01.075

129. Addison NV. Torsion of a mesenteric lymphatic cyst. Br Med J. 1953;1(4823):1316. Available from: https://doi.org/10.1136/bmj.1.4823.1316

130. Chaney JV, Bower RJ, Kudryk B, Gilbert-Barness E. Pathological case of the month. Mesenteric cystic lymphangioma. Arch Pediatr Adolesc Med. 1994;148(8):837-8. Available from: https://doi.org/10.1001/archpedi.1994.02170080067013

131. de Vries JJ, Vogten JM, de Bruin PC, Boerma D, van de Pavoordt HD, Hagendoorn J. Mesenterical lymphangiomatosis causing volvulus and intestinal obstruction. Lymphat Res Biol. 2007;5(4):269-73. Available from: https://doi.org/10.1089/lrb.2007.1010

132. Hatten MT, Hamrick-Turner JE, Smith DB. Mesenteric cystic lymphangioma: radiologic appearance mimicking cystic teratoma. Pediatr Radiol. 1996;26(7):458-60. Available from: https://doi.org/10.1007/bf01377201

133. Khattala K, Rami M, Elmadi A, Mahmoudi A, Bouabdallah Y. Giant cystic lymphangioma of the small bowel mesentery: case report. Pan Afr Med J. 2011;9:46. Available from: https://doi.org/10.4314/pamj.v9i1.71228

134. Levene M, Walker PA, White TA. Mesenteric lymphangioma as a cause of acute abdominal symptoms. Arch Dis Child. 1956;31(160):502-5. Available from: https://doi.org/10.1136/adc.31.160.502

135. Mostofian E, Ornvold K, Latchaw L, Harris RD. Prenatal sonographic diagnosis of abdominal mesenteric lymphangioma. J Ultrasound Med. 2004;23(1):129-32. Available from: https://doi.org/10.7863/jum.2004.23.1.129

136. Mullett JH, Rahim A, Hegarty J, Sheehan K, Murphy JJ. Case report: mesenteric lymphangiomatous cyst. Ir J Med Sci. 1996;165(4):299. Available from: https://doi.org/10.1007/bf02943097

137. Patoulias I, Plikaditi T, Feidantsis T, Ioannidou D, Patoulias D. Clinical image. Chylolymphatic mesenteric cyst in a 3-month-old infant. Folia Med Cracov. 2020;60(1):97-101. Available from: https://doi.org/10.24425/fmc.2020.133490

138. Prabhakaran K, Patankar JZ, Loh DL, Ahamed Faiz Ali MA. Cystic lymphangioma of the mesentery causing intestinal obstruction. Singapore Med J. 2007;48(10):e265-7. Available from: https://pubmed.ncbi.nlm.nih.gov/17909661/

139. Rami M, Mahmoudi A, El Madi A, Khalid, Khattala, Afifi MA, et al. Giant cystic lymphangioma of the mesentery: varied clinical presentation of 3 cases. Pan Afr Med J. 2012;12:7. Available from: https://pubmed.ncbi.nlm.nih.gov/22826732/

140. Steyaert H, Guitard J, Moscovici J, Juricic M, Vaysse P, Juskiewenski S. Abdominal cystic lymphangioma in children: benign lesions that can have a proliferative course. J Pediatr Surg. 1996;31(5):677-80. Available from: https://doi.org/10.1016/s0022-3468(96)90673-9

141. Teixeira L, Castro M, Leite J, Teixeira H, Teixeira R, Pettersen H, et al. Mesenteric cystic lymphangioma: a prenatal diagnostic challenge. Prenat Diagn. 2007;27(5):479-80. Available from: https://doi.org/10.1002/pd.1680

142. Zaki SA, Shenoy P, Shanbag P, Mondkar J, Kalgutkar A. Mesenteric cyst in a neonate causing obstructive uropathy and secondary type 1 pseudohypoaldosteronism - a case report. J Paediatr Child Health. 2011;47(7):486-8. Available from: https://doi.org/10.1111/j.1440-1754.2011.02133.x

143. Elliott GB, Kliman MR, Elliott KA. Persistence of lymphatico-venous shunts at the level of the microcirculation: Their relationship to "lymphangioma" of mesentery. Ann Surg. 1970;172(1):131-6. Available from: https://pubmed.ncbi.nlm.nih.gov/5448503/

144. Esposito C, Alicchio F, Savanelli A, Ascione G, Settimi A. One-trocar ileo-colic resection in a newborn infant with a cystic lymphangioma of the small-bowel mesentery. J Laparoendosc Adv Surg Tech A. 2009;19(3):447-9. Available from: https://doi.org/10.1089/lap.2008.0261

145. López-González Garrido JD, Ramírez-Garrido F, López-González Garrido C, Marín Aznar JL, Valladares Mendías JC, Mingorance MA. Imaging diagnosis of mesenteric cystic lymphangioma: a case report in a newborn. Eur Radiol. 1999;9(4):754. Available from: https://doi.org/10.1007/s003300050752

146. McQuown DS, Fishbein MC, Moran ET, Hoffman RB. Abdominal cystic lymphangiomatosis: report of a case involving the liver and spleen and illustration of two cases with origin in the greater omentum and root of the mesentery. J Clin Ultrasound. 1975;3(4):291-6. Available from: https://doi.org/10.1002/jcu.1870030412

147. Porras-Ramirez G, Hernandez-Herrera MH. Hemorrhage into mesenteric cyst following trauma as a cause of acute abdomen. J Pediatr Surg. 1991;26(7):847-8. Available from: https://doi.org/10.1016/0022-3468(91)90153-k

148. Santo S, Marques JP, Veca P, Melo A, da Graça LM. Prenatal ultrasonographic diagnosis of abdominal cystic lymphangioma: a case report. J Matern Fetal Neonatal Med. 2008;21(8):565-6. Available from: https://doi.org/10.1080/14767050802165927

149. Solari V, Mullassery D, Lansdale N, Jesudason EC. Laparoscopic excision of a retroperitoneal lymphatic malformation in a newborn. J Pediatr Surg. 2011;46(2):e15-7. Available from: https://doi.org/10.1016/j.jpedsurg.2010.09.100

150. Tian C, Zheng Y, Ren X, Li B. A giant abdominal cystic tumor: Mesentery cystic lymphangioma. Dig Liver Dis. 2015;47(9):816-7. Available from: https://doi.org/10.1016/j.dld.2015.05.008

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

151. Vibhav, Amarapurkar AD. Cystic lymphangioma as mesenteric tumor--a case report. Indian J Pathol Microbiol. 2005;48(4):491-3. Available from: https://pubmed.ncbi.nlm.nih.gov/16366106/

152. Yoon HK, Han BK. Chronic midgut volvulus with mesenteric lymphangioma: a case report. Pediatr Radiol. 1998;28(8):611. Available from: https://doi.org/10.1007/s002470050429

153. Amos JA. Multiple lymphatic cysts of the mesentery. Br J Surg. 1959;46:588-92. Available from: https://doi.org/10.1002/bjs.18004620007

154. Fragoyannis SG, Anagnostopulos G. Hemangiolymphomatous hamartoma of the mesentery. Am J Dis Child. 1974;128(2):233-4. Available from: https://doi.org/10.1001/archpedi.1974.02110270107022

155. Humphries PD, Wynne CS, Sebire NJ, Olsen Ø E. Atypical abdominal pediatric lymphangiomatosis: diagnosis aided by diffusion-weighted MRI. Pediatr Radiol. 2006;36(8):857-9. Available from: https://doi.org/10.1007/s00247-006-0173-7

156. Karaca I, Hosgör M, Sencan A, Etensel B, Mir E. Abdominal cystic lymphangioma: a rare cause of acute abdomen in a neonate. Pediatr Int. 2001;43(5):525-6. Available from: https://doi.org/10.1046/j.1442-200x.2001.01428.x

157. Kittredge RD, Finby N. The many facets of lymphangioma. Am J Roentgenol Radium Ther Nucl Med. 1965;95:56-66. Available from: https://doi.org/10.2214/ajr.95.1.56

158. Leonidas JC, Kopel FB, Danese CA. Mesenteric cyst associated with protein loss in the gastrointestinal tract. Study with lymphangiography. Am J Roentgenol Radium Ther Nucl Med. 1971;112(1):150-4. Available from: https://doi.org/10.2214/ajr.112.1.150

159. Neumann DP, Henken EM. Lymphangioma of the jejunal mesentery. J Ultrasound Med. 1997;16(8):563-4. Available from: https://doi.org/10.7863/jum.1997.16.8.563

160. Oshima K, Suzuki N, Ikeda H, Takahashi A, Kuroiwa M, Ohki S, et al. Infected duodenal duplication with unusual clinical and radiological manifestations: a case report. Pediatr Radiol. 1998;28(7):518-20. Available from: https://doi.org/10.1007/s002470050400

161. Radivoyevitch MA, Mirmiran-Yazdy SA. Lymphangiolipoma of the mesentery. Am Surg. 1989;55(7):435-7. Available from: https://pubmed.ncbi.nlm.nih.gov/2662840/

162. Rahman H, Agarwal VK, Gupta SC, Srivastava AK. Lymphangioma of mesentery in a child: a case report. Indian J Pediatr. 1979;46(377):219-21. Available from: https://pubmed.ncbi.nlm.nih.gov/511283/

163. Konen O, Rathaus V, Dlugy E, Freud E, Kessler A, Shapiro M, et al. Childhood abdominal cystic lymphangioma. Pediatr Radiol. 2002;32(2):88-94. Available from: https://doi.org/10.1007/s00247-001-0612-4

164. Merrot T, Chaumoitre K, Simeoni-Alias J, Alessandrini P, Guys JM, Panuel M. Abdominal cystic lymphangiomas in children. Clinical, diagnostic, and therapeutic aspects: apropos of 21 cases. Ann Chir. 1999;53(6):494-9. Available from: https://pubmed.ncbi.nlm.nih.gov/10427841/

165. Perkins JA. New Frontiers in Our Understanding of Lymphatic Malformations of the Head and Neck: Natural History and Basic Research. Otolaryngol Clin North Am. 2018;51(1):147-58. Available from: https://doi.org/10.1016/j.otc.2017.09.002

166. Le Cras TD, Goines J, Lakes N, Pastura P, Hammill AM, Adams DM, et al. Constitutively active PIK3CA mutations are expressed by lymphatic and vascular endothelial cells in capillary lymphatic venous malformation. Angiogenesis. 2020;23(3):425-42. Available from: https://doi.org/10.1007/s10456-020-09722-0

167. Melikhan-Revzin S, Kurolap A, Dagan E, Mory A, Gershoni-Baruch R. A Novel Missense Mutation in FLT4 Causes Autosomal Recessive Hereditary Lymphedema. Lymphat Res Biol. 2015;13(2):107-11. Available from: https://doi.org/10.1089/lrb.2014.0044

168. Niwa H, Sumita N, Ishihara K, Hoshino T, Iwase H, Kuwabara Y. A case of retroperitoneal chylous cyst developed after cholecystectomy and choledochotomy. Nihon Geka Gakkai Zasshi. 1988;89(2):282-5. Available from: https://pubmed.ncbi.nlm.nih.gov/3362131/

169. Gleason TJ, Yuh WT, Tali ET, Harris KG, Mueller DP. Traumatic cervical cystic lymphangioma in an adult. Ann Otol Rhinol Laryngol. 1993;102(7):564-6. Available from: https://doi.org/10.1177/000348949310200714

170. Schmidt M. Intra-abdominal lymphangioma. Kans Med. 1992;93(5):149-50. Available from: https://pubmed.ncbi.nlm.nih.gov/1619838/

171. 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(7):837-40. Available from: https://doi.org/10.1007/s00268-005-7794-0

172. Kosir MA, Sonnino RE, Gauderer MW. Pediatric abdominal lymphangiomas: a plea for early recognition. J Pediatr Surg. 1991;26(11):1309-13. Available from: https://doi.org/10.1016/0022-3468(91)90607-u

173. Adedayo AA, Igashi JB, Mshelbwala PM, Nasir AA, Ameh EA, Adeniran JO. Accuracy of ultrasonography in the evaluation of abdominal masses in children in Nigeria. Afr J Paediatr Surg. 2019;16(1-4). Available from: https://doi.org/10.4103/ajps.ajps_74_16

174. Takiff H, Calabria R, Yin L, Stabile BE. Mesenteric cysts and intra-abdominal cystic lymphangiomas. Arch Surg. 1985;120(11):1266-9. Available from: https://doi.org/10.1001/archsurg.1985.01390350048010

175. Cutillo DP, Swayne LC, Cucco J, Dougan H. CT and MR imaging in cystic abdominal lymphangiomatosis. J Comput Assist Tomogr. 1989;13(3):534-6. Available from: https://doi.org/10.1097/00004728-198905000-00038

176. Mulliken JB, Fishman SJ, Burrows PE. Vascular anomalies. Curr Probl Surg. 2000;37(8):517-84. Available from: https://doi.org/10.1016/s0011-3840(00)80013-1

177. Arraiza M, Metser U, Vajpeyi R, Khalili K, Hanbidge A, Kennedy E, et al. Primary cystic peritoneal masses and mimickers: spectrum of diseases with pathologic correlation. Abdom Imaging. 2015;40(4):875-906. Available from: https://doi.org/10.1007/s00261-014-0250-6

178. Ayyappan AP, Jhaveri KS, Haider MA. Radiological assessment of mesenteric and retroperitoneal cysts in adults: is there a role for chemical shift MRI? Clin Imaging. 2011;35(2):127-32. Available from: https://doi.org/10.1016/j.clinimag.2010.03.003

179. de Perrot M, Bründler M, Tötsch M, Mentha G, Morel P. Mesenteric cysts. Toward less confusion? Dig Surg. 2000;17(4):323-8. Available from: https://doi.org/10.1159/000018872

180. Stopinski J, Stephan S, Staib I. [Intra-abdominal cystic lymphangioma and mesenteric cysts as a cause of abdominal discomfort]. Langenbecks Arch Chir. 1994;379(3):182-7. Available from: https://doi.org/10.1007/bf00680116

181. Okizaki A, Shuke N, Yamamoto W, Usui K, Koyano S, Miyokawa N, et al. Protein-loss into retroperitoneal lymphangioma: demonstration by lymphoscintigraphy and blood-pool scintigraphy with Tc-99m-human serum albumin. Ann Nucl Med. 2000;14(2):131-4. Available from: https://doi.org/10.1007/bf02988593

182. Hancock BJ, St-Vil D, Luks FI, Di Lorenzo M, Blanchard H. Complications of lymphangiomas in children. J Pediatr Surg. 1992;27(2):220-4; discussion 4-6. Available from: https://doi.org/10.1016/0022-3468(92)90316-y

183. Méndez-Gallart R, Bautista A, Estévez E, Rodríguez-Barca P. Abdominal cystic lymphangiomas in pediatrics: surgical approach and outcomes.

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125

Acta Chir Belg. 2011;111(6):374-7.
Available from: https://doi.org/10.1080/00015458.2011.11680776

184. Bertozzi M, Ruffoli M, Vatta F, Gazzaneo M, Raffaele A, Mencherini S, et al. The effectiveness of abdominal lymphangioma laparoscopic removal in children: A single center experience. J Laparoendosc Adv Surg Tech A. 2021;31(12):1367-71.
Available from: https://doi.org/10.1089/lap.2021.0323

185. Vara-Thorbeck C, Toscano Méndez R, Herrainz Hidalgo R, Mata Martín JM, Vara-Thorbeck R. Laparoscopic resection of a giant mesenteric cystic lymphangioma. Eur J Surg (Acta Chirurgica). 1997;163(5):395-6. Available from: https://pubmed.ncbi.nlm.nih.gov/9195174/

186. Luo CC, Huang CS, Chao HC, Chu SM, Hsueh C. Intra-abdominal cystic lymphangiomas in infancy and childhood. Chang Gung Med J. 2004;27(7):509-14.
Available from: https://pubmed.ncbi.nlm.nih.gov/15508873/

Citation: Saxena A, Kakodkar P, Zhang D, Poulin A, Shekari N, Kanthan SC, et al. Intra-Abdominal Multi Cystic Lymphangiomas: A Case Series with Adult and Pediatric Literature Review. Arch Clin Gastroenterol. 2024;10(3):027-039. Available from: https://dx.doi.org/10.17352/2455-2283.000125