MHA MEDIHEALTH ACADEMY

Surgery on Children

Case Report

doi: 10.51271/SOC-0056

Mesenteric lymphatic malformation mimicking chylous ascites: a case report

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Received: 07/04/2025 ◆ **Accepted**: 24/04/2025 ◆ **Published**: 01/10/2025

Cite this article: Altamar MC, Fierro F, Mora N, et al. Mesenteric lymphatic malformation mimicking chylous ascites: a case report. *Surg Child.* 2025;2(4):163-165.

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ABSTRACT

Mesenteric lymphatic malformations (MLM), previously referred to as mesenteric cysts, represent rare clinical entities. While these malformations are often asymptomatic, they can occasionally present with acute abdominal symptoms. The growth patterns as well as the fluid composition may vary. We present a case involving a 5-year-old patient who experienced progressive abdominal distention accompanied by an ascitic wave (initially misdiagnosed as ascites). A paracentesis was conducted, producing a cloudy fluid, which raised concerns for chylous ascites (CA), leading to additional diagnostic assessments. Surgical intervention became necessary when laboratories, fluid analyses, and imaging studies failed to provide a conclusive diagnosis. Ultimately, laparoscopy confirmed the presence of a mesenteric lymphatic malformation. Differentiating between MLM and CA poses significant challenges, highlighting the importance of considering MLM as a crucial differential diagnosis for CA. This paper aims to improve the accuracy of diagnoses to minimize the occurrence of unnecessary interventions. Importantly, the adoption of single-incision laparoscopy has proven to be a safe method, yielding favorable outcomes for this patient. However, continued research is essential to deepen our understanding and establish effective management protocols for these distinctive conditions.

Keywords: Abdominal pain, abdominal distention, mesenteric lymphatic malformations, chylous ascites

INTRODUCTION

Historically, mesenteric cysts and mesenteric lymphangiomas were traditionally described as different entities. Currently, both terms are included in a group called mesenteric lymphatic malformations (MLM).[1] Both entities refer to vascular anomalies rather than lymphatic system tumors.[1,3,4] MLM is rare, occurring in about 1 per 20.000 to 1 per 250.000 hospital admissions,[1,2] with around 5% found in the mesentery, making up 6% of childhood vascular anomalies.[2] These malformations are often asymptomatic,[1,2] but can present with non-specific symptoms or acute abdominal pain.[2,3] There are three described subtypes: microcystic, macrocystic, and mixed.[1] Macrocysts can enlarge, leading to the formation of giant cysts that may resemble ascites.[1] Some of them may contain chylous fluid, warranting differential diagnosis with chylous ascites (CA).[1,2]

CA is the accumulation of chylous-free fluid in the abdominal cavity, typically manifesting as painless abdominal distention but can also include abdominal pain and diarrhea.[5,6] Diagnosis often involves paracentesis showing milky fluid with high triglyceride content (>200 mg/dl), while imaging techniques have limited value.[5,6] Treatment options include a low-fat diet, medications to reduce blood flow, lymphangiography embolism, and surgery.[5,6]

Each entity exhibited varying morbidity and mortality,[2,5,6] highlighting the significance of accurate diagnosis and treatment. This report discusses a case of abdominal distention in a child and outlines the diagnostic process to distinguish between CA and MLM, presenting MLM as a differential diagnosis for CA and its clinical implications.

CASE

A previously healthy 5-year-old male presented to the emergency department with a 1-year history of abdominal

distension. The distension began after a minor abdominal trauma and gradually worsened over time, accompanied by decreased appetite. Upon physical examination, the only notable sign was the presence of an ascitic wave. An initial ultrasound (US) evaluation suggested the presence of an abdominal hematoma. Due to persistent abdominal distension, ascites were suspected, leading to a diagnostic paracentesis that yielded milky fluid. However, further analysis was limited by resources, prompting a referral to our institution. A repeat abdominal US revealed a large volume of fluid displacing adjacent structures without vascular compromise. Laboratory tests, including atrial natriuretic peptide, kidney and liver profiles, serum albumin, and protein levels, were normal, and neoplasm markers (CA 19-9, CA 125, alpha-fetoprotein) were negative.

Subsequently, new paracentesis resulted in the extraction of 2100 ml of milky fluid, with analysis indicating a pH of 8, a leukocyte count of 2993 (92% lymphocytes), 3.5 g/dl albumin, 4.8 g/dl total protein, 49 g/dl glucose, and 960 mg/dl triglycerides. Mycobacterial cultures and cytology were negative. Based on these findings, a diagnosis of CA was established, and the patient was placed on a low-lipid diet with medium-chain triglyceride supplementation. Although intestinal lymphangiectasia was initially suspected as a potential cause of the CA, this was ruled out through upper and lower gastrointestinal endoscopy and histopathological examinations. Lymphangiography was considered but ultimately not conducted due to technical constraints.

A magnetic resonance image (MRI) was performed, and upon initial review, the findings appeared comparable to those observed in the US. However, a more detailed analysis revealed a significant accumulation of non-free abdominal fluid. This fluid was not only a noteworthy detail but also influenced the positioning of the bowel loops, causing them to shift medially (**Figure 1**). This led to the consideration of mesenteric lymphatic malformation vs CA, prompting surgical exploration. During laparoscopic examination, a giant cystic malformation in the ileal mesentery was identified (**Figure 2**). Due to its adherence to the mesentery and compression of the intestinal vasculature, segmental intestinal resection with end-to-end anastomosis was performed.

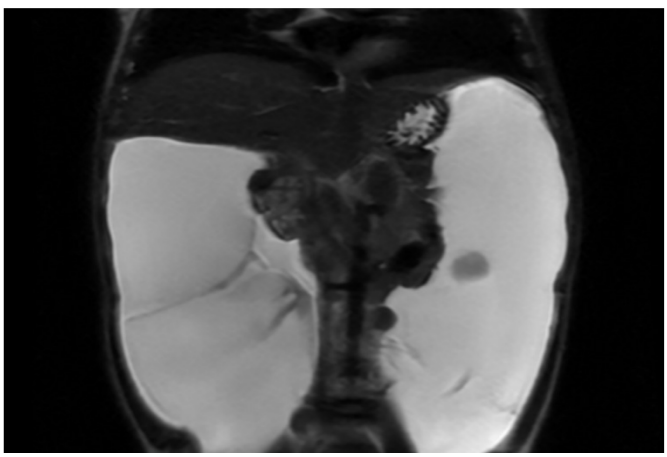

Figure 1. Magnetic resonance image of the patient. The coronal section illustrates the compressive effect of the bowel loops, indicated by the red arrow, which shows medial displacement. The black arrow highlights the trabeculated non-free fluid.

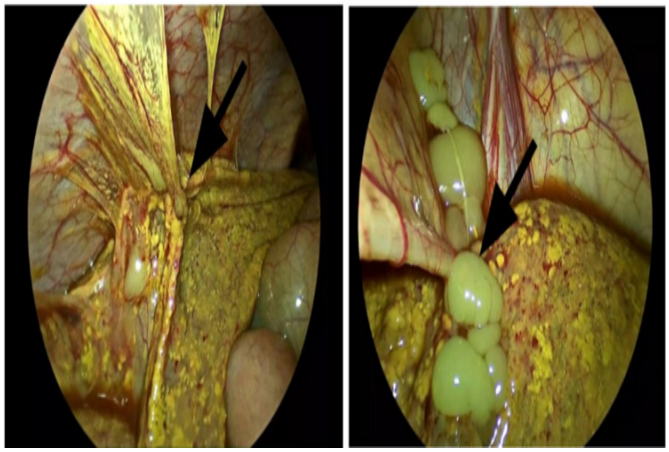

Figure 2. Laparoscopic findings. The left image displays the cyst, indicated by the arrow. The right image reveals the chylous content of the cyst being externalized into the abdominal cavity, as marked by the arrow.

Additionally, 600 ml of chylous fluid was drained from the cyst. The anatomopathological analysis confirmed the presence of a benign cystic lymphatic lesion with extensive xanthogranulomatous inflammation (**Figure 3**). Immunohistochemical features supported the diagnosis of a mesenteric lymphatic malformation. The postoperative period was uneventful, and the patient was discharged five days later.

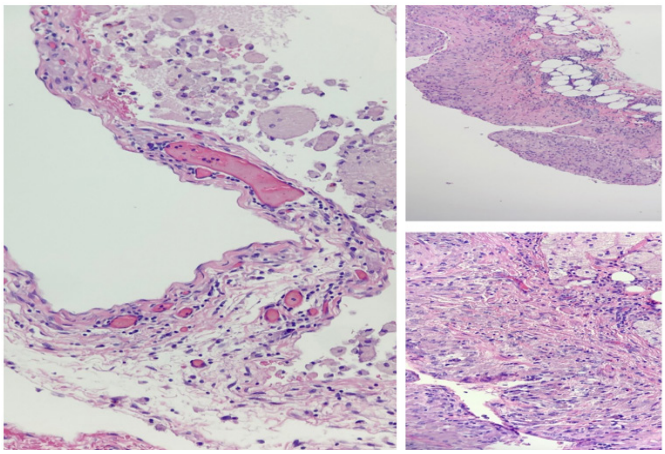

Figure 3. Histology of mesenteric malformations: The sections reveal fibro-adipose tissue with dilated cystic spaces partially covered by endothelial cells devoid of atypia. Additionally, foamy histiocytes are present, along with some lymphoid aggregates and nodules of reactive fibroblastic proliferation.

DISCUSSION

Lymphatic malformations are mainly found in the axilla and neck,[7,8] with the mesentery being a rare location (5% of cases).[1,3,9] The pathophysiology of this entity is still a controversial topic, and consensus has not been reached yet. Theories suggest either abnormal development of lymph vessels or obstruction of lymphatic flow.[1,7,8] Histologically, MLMs feature variable caliber vascular spaces with a protein meshwork, thin endothelial walls, and significant lymphocytic inflammation accompanied by a xanthogranulomatous reaction.[4] While often incidental, MLM can cause symptoms like abdominal pain, distention, nausea, vomiting, constipation, and diarrhea.[2,4] Complications such as hemorrhage, infection, cyst rupture, and volvulus have been noted. Diagnosis typically involves

US, with computed tomography (CT) and MRI offering better cyst characterization.[4,8] There are no established management algorithms; however, surgical excision is preferred for macrocysts, with sclerotherapy and sirolimus also accepted.[9-11]

In this case study, the patient was initially diagnosed with CA based on fluid triglyceride levels (>200mg/dl).[5,6] However, the paracentesis likely sampled fluid from the MLM rather than from free abdominal fluid. Various types of fluids, such as hematic, bilious, or serum, have been described[1,4,8] and chylous fluid in MLM has also been noted.[1,8] However, a large MLM resembling CA has not been documented. The patient's history of low-height abdominal trauma raises suspicion of CA, but CA is typically associated with high-energy trauma.[5]

Our case aligns with the typical epidemiology of MLM, where at least 60-90% are diagnosed by the patient's age,[1-3] with a reported male predominance.[3] Some studies, however, have noted an equal or female distribution.[1,7] The lymphatic malformation in our study was located in the ileal mesentery, a common site for this condition.[7] For symptomatic cases, CA typically presents with abdominal distention,[5,6] while MLM is often associated with abdominal pain.[7] In our patient, the progressive abdominal distention supports a CA diagnosis, although this symptom's frequency may be underestimated in MLM due to its growth alongside the patient,[7] making changes in abdominal perimeter less noticeable.

If well in our diagnostic workup US was the initial study, it was not until MRI was reviewed that the MLM diagnosis was contemplated. This imaging modality differentiates a cystic lesion from free abdominal fluid. Our experience is insufficient to recommend MRI as a diagnostic image in MLM, but it could be helpful to distinguish giant cystic lymphatic malformations from ascitic fluid. Some authors even report MRI as the gold standard for the lymphatic malformations diagnosis.[2] Moreover, it is known that MRI is associated with higher costs, and it is not widely available.

This report discusses a case where complete cyst excision with segmental intestinal resection was performed using single-incision laparoscopy, resulting in no complications during or after the procedure. While surgery remains the main treatment for MLM (especially macrocystic lesions),[7,9] some pediatric surgeons prefer open methods. Nevertheless, single-incision laparoscopy has shown to be a safe alternative, although more studies are needed to confirm its efficacy.[10,11]

CONCLUSION

CA and MLM are distinct yet often confounding conditions due to their overlapping symptoms. It is crucial to recognize each as a potential differential diagnosis for the other. Advanced diagnostic imaging plays a pivotal role in this differentiation; while US may have its limitations, MRI offers significant insights that can effectively distinguish between the two. Including MLM in the differential diagnosis of CA is not just prudent but essential—it can prevent unnecessary treatments and complications, paving the way for timely and effective management of MLM. Furthermore, the adoption of minimally invasive techniques, such as single-incision laparoscopy, represents a promising approach that can lead to favorable patient outcomes. However, further studies that provide higher levels of evidence should be conducted to develop comprehensive recommendations about the role of MRI as a crucial diagnostic tool as well as support the adoption of single-incision laparoscopy as a treatment approach for MLM.

ETHICAL DECLARATIONS

Informed Consent

The patient's parents signed the informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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