


Case Report

Understanding acute chyloperitoneum in appendicitis: case report and literature review

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Abstract

Chyloperitoneum is the presence of chyle in the peritoneal cavity. This study focuses on acute chyloperitoneum, a rare condition with an unclear incidence due to limited number of reported cases in the literature. Here, we present a 24-year-old Saudi female with chyloperitoneum diagnosed intraoperatively during a laparoscopic appendectomy for acute appendicitis that was managed successfully with a low-fat diet and drainage, alongside a literature review to elucidate the condition's pathophysiology and therapeutic strategies. A conservative management approach is recommended for acute chyloperitoneum in the context of appendicitis, this includes intraperitoneal drainage, appendectomy when needed, and careful observation. Our proposed management strategy aligns with findings from the literature review and supports conservative management as a safe and effective treatment modality for this rare condition.

Keywords: chyloperitoneum; chyle Leak; chylous ascites; appendicitis

Introduction

Chyloperitoneum, also known as chylous ascites, is the abnormal accumulation of chyle in the peritoneal cavity, which normally circulates through the lymphatic system.

Chyle is consisted of protein, lymphocytes, immunoglobulins, and chylomicrons. Long-chain triglycerides are broken down into monoglycerides and free fatty acids, which are then absorbed in the gut as chylomicrons [1]. The high triglyceride content in chyle gives it a milky and cloudy appearance. A triglyceride level exceeding 200 mg/dL in the ascitic fluid analysis is a key diagnostic indicator for chylous ascites [2].

Causes can be congenital, such as in cases of lymphangiomas, Turner syndrome, and Noonan syndrome due to lymphatic malformations, typically seen in children. They can also be acquired, more common in adults, due to factors such as lymphoma, iatrogenic injury, trauma, chronic peritoneal dialysis, or various other reasons [3]. In this study, we are focusing on acute chyloperitoneum, a condition with an unclear incidence due to a limited number of reported cases, indicating its rarity.

Management of chyle leak usually follows a step-up approach [4], starting with dietary modifications such as a high-protein, low-fat diet with medium-chain triglycerides. If no improvement is seen, pharmacological agents such as octreotide may be used, along with or without total parenteral nutrition (TPN). If these measures fail, further investigations through lymphoscintigraphy to locate the site of leakage are recommended. Subsequent

management varies based on scan results, underlying causes, and available resources, with options including percutaneous embolization, transjugular intrahepatic portosystemic shunt, peritoneovenous shunting, and surgery. However, there is no standardized approach for incidentally discovered acute chyloperitoneum.

Here, we present a case of acute appendicitis with incidental finding of chyloperitoneum and literature review on the topic aim to explore the pathophysiology and management strategies for chyloperitoneum in the context of appendicitis.

Case report

A 24-year-old, unmarried, Saudi female, presented to the emergency department with severe abdominal pain for 3 days. The pain initially began at the paraumbilical area and subsequently migrated to the right lower quadrant. It was gradual in onset, with no known aggravating or relieving factors. The pain was associated with nausea and anorexia, but with no episodes of vomiting. The patient reported no changes in bowel habits, melena, hematochezia, weight loss, night sweats, fever, recent travel, or trauma. She had no history of similar previous attacks. Her menstrual cycle was regular. The patient denied smoking or alcohol use. She had no significant past medical or surgical history, and her family history was unremarkable.

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Table 1. Laboratory results upon presentation.

Labs	Results	Normal range
WBC	12.5 × 10e3/uL	4.00–11.00
Hb	12.9 g/dL	13.5–17.2
Platelets	274 × 10e3/uL	150–450
Na	139.0 mmol/L	136.0–145.0
K	3.60 mmol/L	3.60–5.00
Amylase	51.00 U/L	25.00–125.00
Lipase	29.0 U/L	8.0–78.0

WBC = White blood cells; Hb = Haemoglobin; Na = Sodium; K = Potassium.

a



b



Figure 1. Preoperative enhanced CT abdomen and pelvis with axial (a) and coronal (b) views showing dilated appendix with hyperdense appendicolith within its lumen (arrow) associated with surrounding fat stranding and moderate amount of free fluid of –7 to 0.4 Hounsfield Units density.

Upon presentation, the patient was vitally stable except for a low-grade fever (37.8°C). Her abdominal examination revealed tenderness in the paraumbilical and right lower quadrant regions with positive rebound and Rovsing signs. Abdominal X-ray was unremarkable. Laboratory investigations revealed leukocytosis (Table 1). Enhanced CT scan of the abdomen and pelvis revealed signs of appendicitis with a moderate amount of free fluid (Fig. 1a and b).

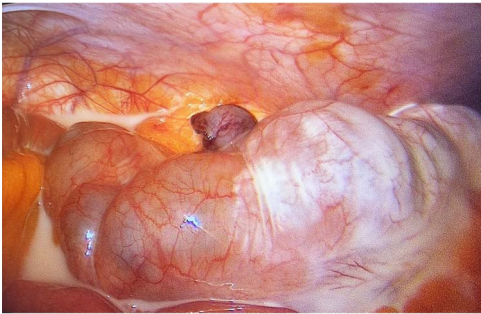


Figure 2. Laparoscopic view of the right lower quadrant showing chyloperitoneum with appendix tip in the middle (post ligation of appendicular artery).

The patient was taken to the operating room for a laparoscopic appendectomy. Intraoperatively, a large amount of milky fluid was discovered in the abdomen along with an inflamed appendix (Fig. 2). The fluid was aspirated and sent for bacterial culture, tuberculosis culture, cytology, and triglyceride level analysis. An attempt to identify the leak site was unsuccessful, so we proceeded with appendectomy and placed a drain in the pelvis. Postoperatively, the patient was transferred to the ward. The triglyceride level in the peritoneal fluid was markedly elevated, with a level of 2029.00 mg/dl, confirming a diagnosis of chylous ascites. All other peritoneal fluid investigations were negative.

The patient was managed with a low-fat diet, octreotide, and close monitoring of drain output. Over the following days, the drain output gradually diminished and became serous in color. By the third postoperative day, the drain was removed, and patient was discharged from the hospital in good condition. During follow-up in the outpatient clinic, the patient reported no active complaints and was in good health. A follow-up enhanced CT scan of the abdomen and pelvis, performed 1 month after discharge, showed no signs of recurrence (Fig. 3a and b). Histopathology results of the removed appendix confirmed the diagnosis of acute appendicitis.

Discussion

Chyloperitoneum is a rare condition characterized by the presence of chyle in the peritoneal cavity. We encountered a case of acute appendicitis with accumulation of chyle in the abdomen, and the underlying cause of this incidental finding is unclear. The principal mechanisms behind chyloperitoneum as described by Aalami et al. [3] involve the exudation of chyle from dilated lymphatics on the bowel wall and within the mesentery due to lymphatics obstruction in the mesentery or cisterna chyli.

Our hypothesis behind chyloperitoneum in appendicitis is that severe inflammation may damage or obstruct chyle flow in the surrounding mesentery, leading to exudation of chyle and causing its accumulation in the peritoneal cavity (Fig. 4).

In reviewing the literature, similar cases were examined, analyzed, and summarized in Table 2; the mean age of patients was 33.6 years (ranging from 6 to 69 years). Males predominated with a male-to-female ratio of 3:2; majority of cases reported no history of alcohol or smoking use, and none had a history of trauma.

Most patients had no prior surgeries except for three. One of whom had an appendectomy, while the remaining two had non-abdominal surgeries. Acute right iliac fossa pain was a common symptom in all cases, but only 3 out of 15 had operative findings of appendicitis (20%), with the remaining cases lacked other identifiable pathologies to explain the symptoms.

Table 2. Literature review summary of cases with acute chyloperitoneum.

Author / year	Age	Sex	Smoking	Alcohol use	History of trauma	Presence of appendicitis	Other intraoperative findings	Intervention	Drain removal in days	Use of TPN	Use of octreotide	Recurrence
Fazili <i>et al.</i> (1999) [5]	25	M	Yes	No	No	No	None	Intraperitoneal drainage	2	NA	NA	No recurrence in 9 months follow up
Fang <i>et al.</i> (2006) [6]	22	M	No	No	No	No	Lymphatic drainage from thoracic duct	Suture ligation of lymphatic leakage from thoracic duct and retroperitoneal drainage	NA	No	NA	No recurrence in 6 months follow up
Vettoretto <i>et al.</i> (2008) [7]	69	M	NA	NA	No	-	Small vessel hypertrophy forming little angiomias on the mesenteric side of the bowel	Intraperitoneal drainage	7	Yes	NA	No recurrence in 2 years follow up
Akbulut <i>et al.</i> (2010) [8]	25	M	No	No	No	Yes	Proximal jejunal lymphangiectasia	Appendectomy and intraperitoneal drainage	7	Yes	Yes	No recurrence in 1 month follow up
Rogdakis <i>et al.</i> (2011) [9]	46	M	Yes	Yes	No	No	None	Intraperitoneal drainage	7	Yes	Yes	No recurrence in 6 months follow up
Ozgüçet <i>et al.</i> (2013) [10]	32	F	No	No	No	No	None	Intraperitoneal drainage	7	No	Yes	No recurrence in 9 months follow up
Xu <i>et al.</i> (2015) [11]	6	M	No	No	No	No	None	Prophylactic appendectomy	-	No	No	NA
Ul Ain <i>et al.</i> (2016) [12]	32	F	No	Yes	No	No	None	Prophylactic appendectomy and intraperitoneal drainage	?5	NA	NA	No recurrence in 6 months follow up
Kaya <i>et al.</i> (2017) [13]	30	M	NA	NA	NA	Yes	Edematous small bowel mesentery with enlarged lymphatic vessels	Appendectomy and intraperitoneal drainage	5	Yes	Yes	NA
Alamri <i>et al.</i> (2020) [14]	32	M	NA	NA	No	No	None	Intraperitoneal drainage	7	No	Yes	NA
Manco <i>et al.</i> (2020) [15]	30	F	NA	NA	No	No	None	Prophylactic appendectomy and intraperitoneal drainage	3	NA	NA	No recurrence in 5 months follow up
Apikotoa <i>et al.</i> (2021) [16]	36	F	No	No	No	No	Dilated lymphatic vessels on the surface of the small bowel	Prophylactic appendectomy and intraperitoneal drainage	5	No	No	No recurrence in 2 weeks follow up
Epelde <i>et al.</i> (2024) [17]	35	F	NA	NA	No	No	None	Intraperitoneal drainage	?4	No	Yes	No recurrence in 1 year follow up
Zenati <i>et al.</i> (2024) [18]	61	M	No	No	No	No	Dilated lymphatic vessels on the surface of the small bowel	Intraperitoneal drainage	NA	No	NA	No recurrence in 3 years follow up
Current study	24	F	No	No	No	Yes	None	Appendectomy and intraperitoneal drainage	3	No	Yes	No recurrence in 2 months follow up

M = Male; F = Female; NA = Not available

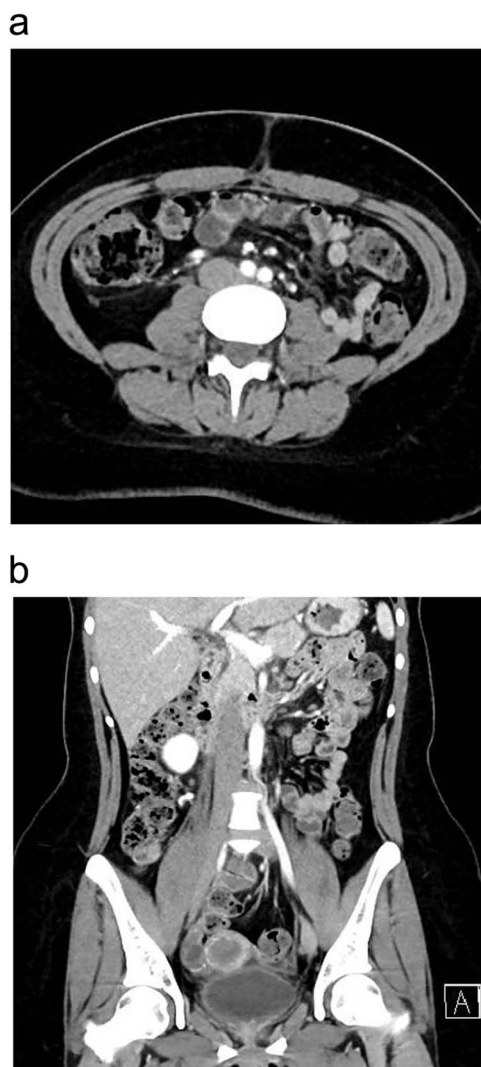


Figure 3. One month postoperatively enhanced CT abdomen and pelvis with axial (a) and coronal (b) views showing resolution of inflammation and no signs of recurrence of chyloperitoneum.

While retroperitoneal exploration was carried out in 3 out of 15 cases, only 1 patient showed lymphatic leakage from thoracic duct and was repaired through suture ligation [6]; all other cases were unable to locate a leak site.

Drainage played a significant role in management, either alone (53.3%) or in conjunction with appendectomy (46.7%). Fortunately, all cases showed no recurrence of chyloperitoneum during follow-up.

In our case and similar cases in the literature, chyle leakage was self-limiting, supporting the theory that it is linked to mesenteric inflammation, and resolution of the inflammation led to a decrease in the rate of chyle leakage and eventual cessation.

Based on our review of the literature and our own experience, we suggest adopting a conservative approach when managing acute chyloperitoneum associated with appendicitis. Avoiding aggressive interventions is key to minimizing complications (Fig. 5).

Intraperitoneal drainage, along with appendectomy if indicated, accompanied by observation is sufficient as demonstrated by our findings and similar cases in the literature, unless a specific leakage site or another underlying pathology is confirmed.

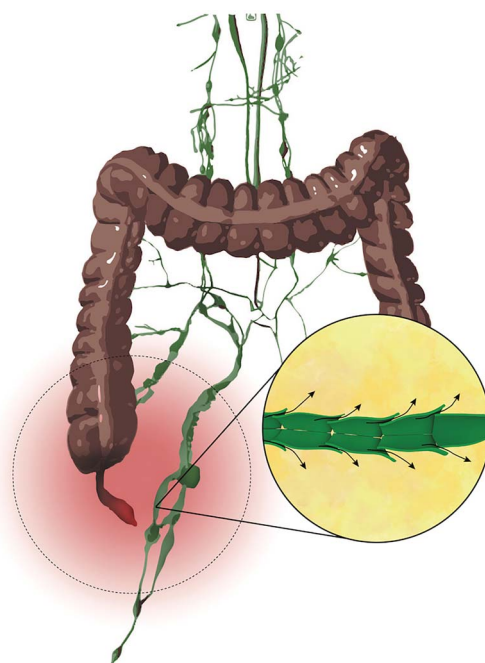


Figure 4. Illustration of the hypothetical pathophysiology behind acute chyloperitoneum with appendicitis.

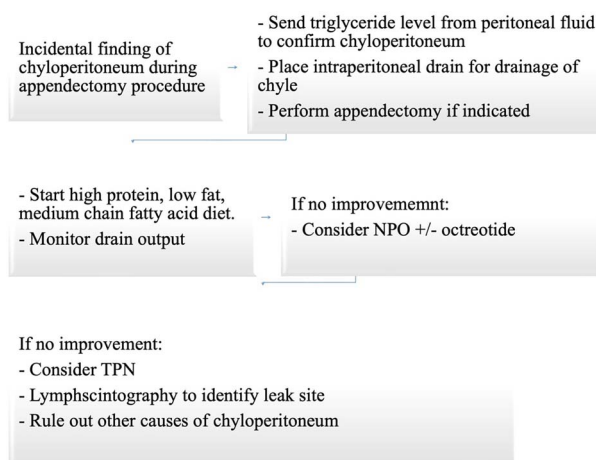


Figure 5. Suggested approach for incidental finding of chyloperitoneum intraoperatively during appendectomy.

Conclusion

Although there is no established relationship between appendicitis and chyloperitoneum, we hypothesize that surrounding mesenteric inflammation may be a connecting factor.

Given the lack of a standardized treatment approach in the literature for incidentally discovered acute chyloperitoneum during an appendectomy, a conservative management approach is advised in such cases. This approach includes intraperitoneal drainage, appendectomy if needed, and careful observation. Our proposed management strategy aligns with findings from the literature review and supports conservative management as a safe and effective treatment modality for this rare condition.

Further studies are needed to elucidate how appendicitis could be related to chyloperitoneum, to guide diagnostic and therapeutic strategies to effectively manage this condition.

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Conflict of interest statement

None declared.

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References

1. Al-Busafi SA, Ghali P, Deschênes M, et al. Chylous ascites: evaluation and management. *ISRN Hepatology* 2014;**2014**: 240473.
2. Lizaola B, Bonder A, Trivedi HD, et al. Review article: the diagnostic approach and current management of chylous ascites. *Aliment Pharmacol Ther* 2017;**46**:816–24. <https://doi.org/10.1111/apt.14284>.
3. Aalami OO, Allen DB, Organ CH Jr. Chylous ascites: a collective review. *Surgery* 2000;**128**:761–78. <https://doi.org/10.1067/msy.2000.109502>.
4. Bhardwaj R, Vaziri H, Gautam A, et al. Chylous ascites: a review of pathogenesis, diagnosis and treatment. *J Clin Transl Hepatol* 2018;**6**:105–13. <https://doi.org/10.14218/JCTH.2017.00035>.
5. Fazili FM, Khawaja FI. Acute chylous peritonitis simulating acute appendicitis: a case report and review of the literature. *Ann Saudi Med* 1999;**19**:236–8. <https://doi.org/10.5144/0256-4947.1999.236>.
6. Fang FC, Hsu SD, Chen CW, et al. Spontaneous chylous peritonitis mimicking acute appendicitis: a case report and review of literature. *World J Gastroenterol* 2006;**12**:154–6. <https://doi.org/10.3748/wjg.v12.i1.154>.
7. Vettoretto N, Odeh M, Romessis M, et al. Acute abdomen from chylous peritonitis: a surgical diagnosis. Case report and literature review. *Eur Surg Res* 2008;**41**:54–7. <https://doi.org/10.1159/000129599>.
8. Akbulut S, Yılmaz D, Bakir S, et al. Acute appendicitis together with chylous ascites: is it a coincidence? *Case Rep Med* 2010;**2010**:206860. <https://doi.org/10.1155/2010/206860>.
9. Rogdakis A, Bouras P, Giannakakis P, et al. Spontaneous chylous peritonitis presenting as acute surgical abdomen. *Hellenic J Surg* 2011;**83**:166–9. <https://doi.org/10.1007/s13126-011-0022-5>.
10. Özgüç H, Narmanlı M, Keskin MK. Acute chylous peritonitis: report of a case. *Int J Surg Case Rep* 2013;**4**:419–21. <https://doi.org/10.1016/j.ijscr.2013.01.022>.
11. Xu J, Nair R. Acute chylous ascites mimicking appendicitis. *ANZ J Surg* 2017;**87**:737–9. <https://doi.org/10.1111/ans.12999>.
12. Ul Ain Q, Bashir Y, Johnston S. Idiopathic chylous ascites simulating acute appendicitis: a case report and literature review. *Int J Surg Case Rep* 2016;**29**:189–92. <https://doi.org/10.1016/j.ijscr.2016.10.059>.
13. Kaya C, Yazıcı P, Kartal K, et al. A rare cause of acute abdomen: Chylous ascites. *Turk J Surg* 2015;**33**:123–5. <https://doi.org/10.5152/UCD.2015.2920>.
14. Alamri MS, Alenezi RA, Alanazi KK, et al. Acute chylous peritonitis: report of a case and literature review. *Saudi Surg J* 2020;**8**: 101–4. https://doi.org/10.4103/ssj.ss_j_5_19.
15. Manco G, Caramaschi S, Prestigiacomo G, et al. Idiopathic chylous peritonitis mimicking acute appendicitis a case report. *Ann Ital Chir* 2021;**10**:S2239253X21033880.
16. Apikotoa S, Wijesuriya R. Idiopathic acute chylous peritonitis during pregnancy, mimicking perforated acute appendicitis: a case report. *Int J Surg Case Rep* 2021;**81**:105790. <https://doi.org/10.1016/j.ijscr.2021.105790>.
17. Epelde C, Saravia F, Aguinaga M, et al. Acute abdomen in pregnancy due to idiopathic Chylous ascites. *Hindawi Case Rep Obstet gynecol* 2024;**2024**:1–5. <https://doi.org/10.1155/2024/8898451>.
18. Zenati H, Jallali M, Korbi A, et al. Acute primary chylous peritonitis mimicking acute abdomen: a case report and literature review. *Pan African Medical Journal* 2024;**47**:131. <https://doi.org/10.11604/pamj.2024.47.131.42794>.