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Idiopathic chylous ascites simulating acute appendicitis: A case report and literature review

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1. Introduction

Chylous ascites is the extravasation of milky chyle into the peritoneal cavity. Though formation of chylous ascites is a chronic process, rarely, there can be a sudden release of chyle into the peritoneal cavity, presenting as an acute abdomen. Management mainly involves identifying the cause and treating it. Very few cases of acute chylous peritonitis have been described in literature [1]. Considering its rarity and striking resemblance to common surgical emergency such as acute appendicitis, we present and discuss the management of acute chylous ascites. This is reported in accordance with the SCARE criteria [2].

2. Case report

A 32-year-old female was referred by family physician to the emergency department in secondary care hospital with three days history of lower abdominal pain and nausea. The pain had initially started as dull ache in the lower abdomen but later increased in severity and localized at right lower quadrant. The pain was continuous, severe and aggravated on movement. There was no previous history of trauma. She did not suffer from anorexia, vomiting or altered bowel habits. She had regular menstrual cycle. In the past medical history, patient suffered with depression and CIN3 (cervical intraepithelial neoplasia) and underwent LETZ therapy, three years ago, with no recurrence. She was currently on Escitalopram (SSRIs) once daily for mild depression and oral contraceptive pills that she had stopped three weeks ago. She had no drug allergies. She was a non-smoker and consumes alcohol occasionally. There

is no history of diabetes mellitus, hypertension, hyperlipidemia or recurrent abdominal pains in her or her family.

On examination, she was vitally stable and had an unremarkable systemic examination. The abdomen was soft and ovoid in shape and there was tenderness at lower abdomen (more on right iliac fossa as compared to left) with positive rebound tenderness. Bowel sounds were audible. Blood analysis revealed elevated white cell count (WCC) with neutrophilia and normal liver, renal and lipid profile. (Hb-13.2 g/dL, WCC-13.1 × 10⁹/L [9], Neutrophils-11.41 × 10⁹/L, lymphocytes-1.11 × 10⁹/L [9]. Platelets-230 × 10⁹/L, Urea-4.4 mmol/L, Creatinine-67 mmol/L, Sodium-141 mmol/L, Potassium-4.2 mmol/L, AST-19U/L, ALT-15U/L, ALP-79U/L, Amylase-44U/L).

Her urine analysis, chest x-ray and abdominal x-ray were normal and pregnancy test was negative.

With the above clinical and biochemical findings, acute appendicitis was suspected (Alvarado's score of 7) and senior registrar planned laparoscopic appendectomy for the patient due to acute abdomen findings.

When entering the peritoneum, we noticed moderate amount of white, odorless, non-clotting ascitic fluid in the perihepatic region and pelvis. (Figs. 1 and 2) The Appendix was grossly normal. There was no abnormality visible in both solid and hollow abdominal viscera, no obvious signs of injury to thoracic duct and no dilated lymphatics. Samples of ascitic fluid were taken for bacteriological and biochemical examination. A provisional diagnosis of chylous ascites was made and prophylactic appendectomy performed. A thorough peritoneal wash was performed with 2L of warm saline and port site incisions closed.

Biochemical analysis of the peritoneal fluid revealed isolated high triglycerides, 21 times higher than plasma, confirming it to be chyle. (glucose 2.1 mmol/L, LDH 7.10U/L, triglycerides-21 mmol/L, protein-18.7 g/L, albumin-11.8 g/L. Simultaneous plasma protein level was 60.9 g/L, albumin-38.3 g/L, plasma triglyceride level-0.86 mmol/L. serum amylase was normal). The fluid was bac-

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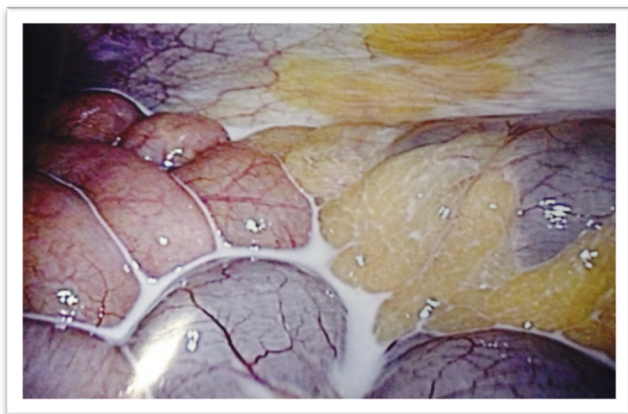


Fig. 1. Chylous ascites in pelvis.

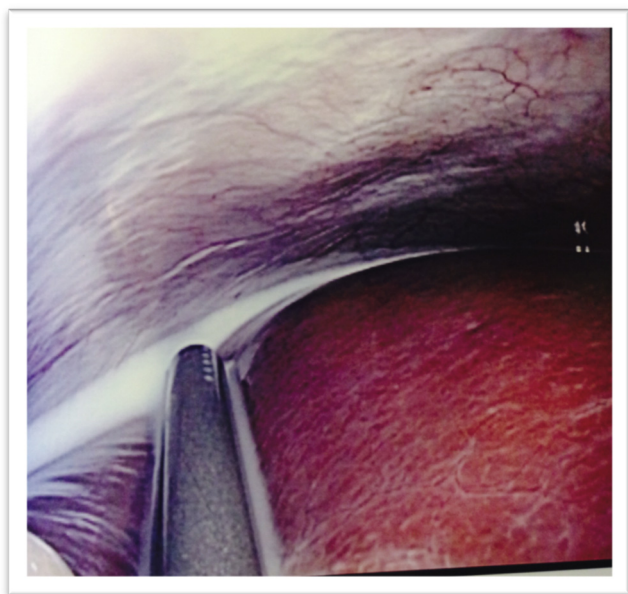


Fig. 2. Chylous ascites in perihepatic region.

terologically sterile and negative for ZN staining. There were no malignant cells seen in cytology.

Post operatively; patient had CT scan of thorax, abdomen and pelvis, which revealed moderate bibasal pleural effusion most likely chylothorax and basal

collapse. The remainder of examination was unremarkable. She also had her serum tumor markers and filariasis screening done, which were both negative. (Fig. 3) Postoperative period was uneventful and patient was discharged on 5th postoperative day with diagnosis of idiopathic acute chylous peritonitis. She was advised to have low fat diet. She remained in good general health and further 6-weekly and 6-monthly follow up did not reveal any malignancy or recurrence of chylous ascites/chylothorax.

3. Discussion

3.1. Introduction

The word, Chyle, or chylaskos has been derived from the Greek word meaning 'juice'. It is rich in triglycerides, as well as protein, electrolytes, lymphocytes, and other substances.

The incidence of chyle leak into peritoneum, i.e. chylous ascites, occurs one in 20,000 patient admitted to hospital [1]. Morton rec-

ognized it for the first time, in 1691 [3] whereas Renner identified the first acute case in 1910 [4]. Since then, very few cases have been reported in literature and little information is available.

3.2. Pathophysiology

Various mechanisms have been described for chylous ascites. Andreys et al [3]. believed that chylous ascites occurred either due to primary fibrosis of lymph nodes (as in malignancy), extravasation of lymph through walls of retroperitoneal lymphatic system (both by congenital malformation or trauma) or due to cardiovascular disorders [5]. Krizek et al [6], however, identified acute cases and classified them into traumatic, obstructive, due to mesenteric cysts or idiopathic.

3.3. Causes

Presence of chylous ascites can occur due to multiple etiologies that can be broadly classified as traumatic and atraumatic.

In a recent systematic review including 131 studies from developing and developed countries, with a total of 190 patients, who had atraumatic chylous ascites, the most common causes in adults were malignancy (25%), cirrhosis (16%) and mycobacterium infection (15%). In children, the most common cause was lymphatic anomalies (84%). Furthermore, Abdominal malignancy and cirrhosis are the commonest causes in developed countries and account for over two-thirds of all cases, whereas majority of cases in developing countries were chronic infections like tuberculosis and filariasis [7] (Table 1).

Among the traumatic causes, abdominal surgery for abdominal aortic aneurysm or retroperitoneal lymph node dissection is the most common etiology [8].

While literature review shows few reported cases of chylous ascites with chylothorax, most of them were due to ongoing chronic illnesses and did not have an acute presentation as opposed to our case, making the reports irrelevant to our case [9,10].

3.4. Clinical features

Painless, progressive abdominal distention (81%) and non-specific pain (14%) are the most common presenting symptoms, which occurs over weeks to months [7]. An acute presentation is less common due to the fact that chyle is non-irritating to the serosal surface and pain is mainly due to stretching of retroperitoneum and mesenteric serosa [11].

Due to increase in abdominal girth, weight gain and dyspnea can also be the primary complaint [3]. Other features include weight loss, anorexia, malaise, steatorrhea, malnutrition, enlarged lymph nodes, fevers, and night sweats [1]. However due to non-specific symptoms, the diagnosis is rarely made before diagnostic paracentesis. Physical signs that may be present on examination include ascites, pleural effusions, lower extremity edema, lymphadenopathy, cachexia, temporal wasting, hernias and findings related to underlying cause [3].

In addition, loss of chyle may result in complications because of loss of essential proteins, lipids, immunoglobulins, vitamins, electrolytes, and water. These complications range from immunosuppression secondary to hypogammaglobulinemia, nutritional deficiency and sub therapeutic levels of various drugs including digoxin and amiodarone.

3.5. Diagnosis

Due to the ambiguous presentation of chylous ascites, diagnosis is usually made on abdominal paracentesis. Its chief characteristics include milky appearance, alkaline chemical properties, specific

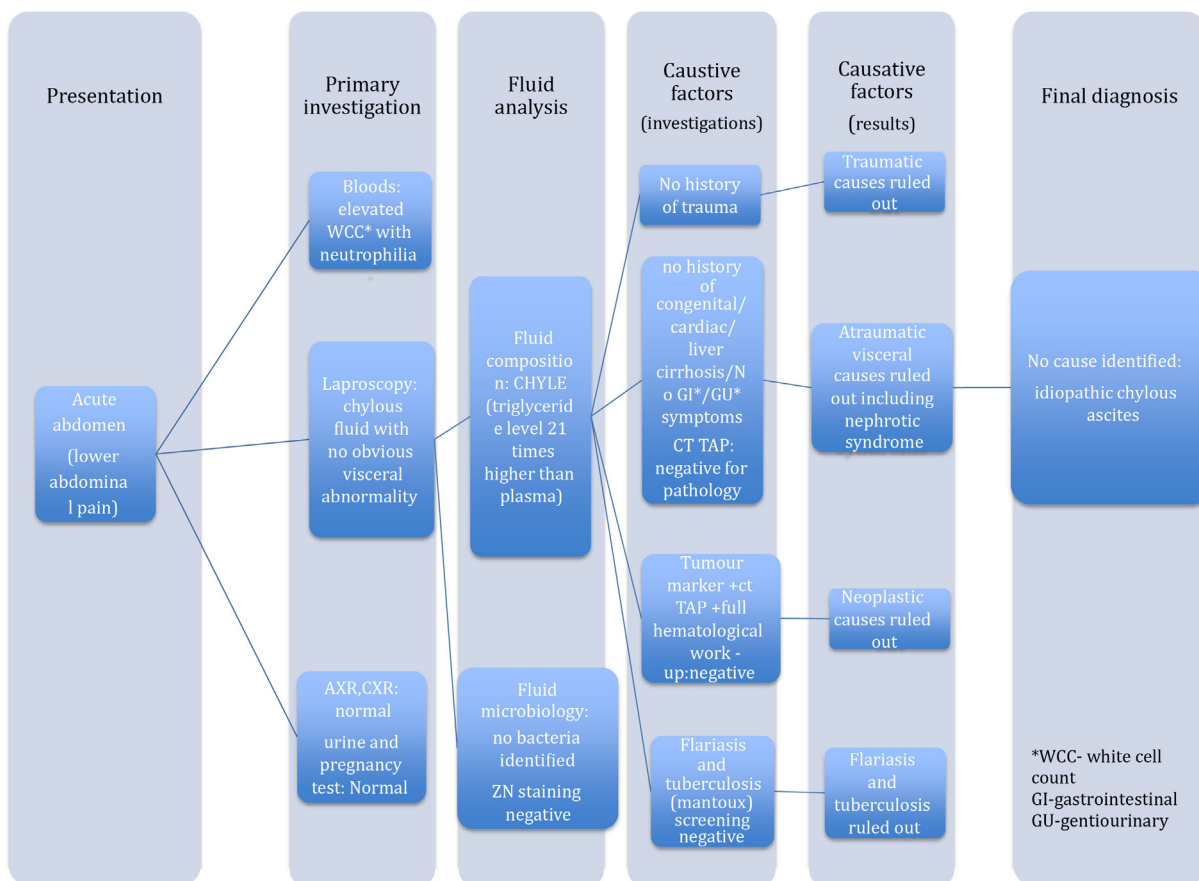


Fig. 3. Investigations for determining chylous ascites in patient.

Table 1 Etiological classification of chylous ascites.

ATRAUMATIC		TRAUMATIC
(I) Neoplastic	Cardiac	(I) Iatrogenic
Solid organ cancers	Constrictive pericarditis	(A) Surgical
Lymphoma	Congestive heart failure	Abdominal aneurysm repair
Sarcoma	Gastrointestinal	Retroperitoneal lymphadenectomy
Carcinoid tumors	Celiac sprue	Placement of peritoneal dialysis catheter
Lymphangioliomyomatosis	Whipple's disease	Inferior vena cava resection
Chronic lymphatic leukemia	Intestinal malrotation	Pancreaticoduodenectomy
(II) Diseases	Small bowel volvulus	Vagotomy
(A) Congenital	Ménétrier disease	Radical and laparoscopic nephrectomy
Primary lymphatic hypoplasia	Inflammatory	Nissen fundoplication
Klippel-Trenaunay syndrome	Pancreatitis	Distal splenorenal shunts
Yellow nail syndrome	Fibrosing mesenteritis	Laparoscopic adrenalectomy
Primary lymphatic hyperplasia	Retroperitoneal fibrosis	Gynecological surgery
Lymphangioma	Sarcoidosis	(B) Nonsurgical
Familial visceral myopathy	Systemic lupus erythematosus	Radiotherapy
(B) Acquired	Behçet's disease	(II) Noniatrogenic
Cirrhosis	Peritoneal dialysis	Blunt abdominal trauma
Infectious	Hypert thyroidism	Battered child syndrome
Tuberculosis	Nephrotic syndrome	Penetrating abdominal trauma
Filarisis	Drugs	Shear forces to the root of the mesentery
Mycobacterium avium in AIDS	Calcium channel blockers	(III) Idiopathic
Ascariasis	Sirolimus	Rule out lymphoma

gravity greater than 1.012, stain positive for fat (Sudan III) and ether test, bacteriostatic in nature, sterile in culture and resisting putrefaction [12]. It is odorless, and only occasionally smells like digested food [13]. Its chemical composition is quite specific. Only chyle has fat content greater than plasma (0.04%–4%). It has protein level of 2.5–7.0 g/dL, SAAG less than 1.1 (except in cirrhosis), total solids >4%, cell count of above 500 (lymphocytic predominance) [14].

The triglyceride level is an important diagnostic tool, and the concentration in chylous ascites is typically 2–8 times that of plasma [15].

Depending on the clinical suspicion, ascitic fluid should be sent for cell count, culture, Gram stain, glucose, lactate dehydrogenase, amylase, and cytology [16]. A tuberculosis smear and culture and adenosine deaminase activity (ADA) should be performed in

selected cases when tuberculosis is suspected [3]. And routine blood tests should be conducted as well.

Other diagnostic studies such as computed tomography, lymphangiography, lymphoscintigraphy and laparotomy have the high yield of diagnostic information [3,17].

3.6. Management

The optimal management of true chylous peritonitis depends upon underlying etiology. In acute cases, urgent exploration is mandatory. This provides definitive diagnosis and correction of cause both laparoscopically or through microsurgical techniques [18]. When a specific cause is not identified at the time of surgery, retroperitoneal dissection is not advisable as spontaneous closure of leak and complete recovery is the rule, as occurred in our case [13].

The aim for non-surgical treatment is to reduce chyle production and improve nutrition. These include administration of total parenteral nutrition, high protein and low fat diet, restrict fats to medium chain triglyceride. Somatostatin improves chylous ascites by inhibiting lymph fluid excretion through specific receptors found in wall of lymphatic vessels [19]. If patients are poor candidate for surgical intervention and refractory to nonsurgical management, peritoneovenous shunting may be an option [20].

4. Conclusion

In summary, we have described a rare case of acute chylous peritonitis that mimicked acute appendicitis, with no identifiable etiology, making it idiopathic. Laparoscopy with peritoneal lavage resulted in complete recovery of the patient. This case shows the effectiveness of laparoscopy in both the diagnosis and treatment of acute abdominal pathology of unknown origin. It also establishes the necessity for further studies to assess and develop management protocols for such conditions.

Conflict of interest

There are no conflicts of interest or financial acknowledgment in this case report.

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Ethical approval

Ethical committee approval not required for this case report.

Consent

Consent taken from patient. And it can be presented to editor whenever necessary.

Author contribution

Qurat ul ain sumra: writing the manuscript.

Yasir bashir: writing the manuscript.

Sean Johnston: reviewer.

Guarantor

Qurat ul ain sumra.

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