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Case Report

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Chylous ascites as a diagnostic surprise during inguinal hernia surgery: a case report and review of literature

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ABSTRACT

Chylous ascites is a rare form of ascites characterized by the accumulation of lipid-rich lymphatic fluid in the peritoneal cavity. In children, congenital lymphovascular malformations like cystic lymphangioma and mesenteric cyst are the most common cause. However, its intraoperative detection during elective procedures like inguinal hernia repair is exceedingly rare. We report a case of 5-month-old male infant presented with reducible right inguinal hernia. During elective herniotomy milky white fluid was encountered, suspicious of chylous ascites. Fluid analysis confirmed the diagnosis with triglyceride levels >9500 mg/dl and positive chylomicrons. Subsequent imaging revealed a cystic lesion in the mesentery and the child underwent exploratory laparotomy with complete excision of the lesion and bowel anastomosis. Histopathology confirmed cystic lymphangioma. Postoperatively the child remain stable and tolerated orals well. At one-year follow-up, the child remains asymptomatic and thriving well. This case highlights the importance of maintaining a high index of suspicion for chylous ascites when atypical intra-abdominal fluid is encountered during surgery. Ruptured cystic lymphangioma presenting as chylous ascites encountered during hernia surgery is a very rare phenomena. With prompt evaluation and appropriate surgical management based on the underlying etiology we can achieve excellent outcomes. Reporting such rare intraoperative findings adds valuable insight into the etiological spectrum and surgical management of pediatric chylous ascites.

Keywords: Chylous ascites, cystic, hernia, inguinal, infant, lymphangioma

INTRODUCTION

Extravasation and accumulation of lymphatic fluid in the abdominal cavity is referred as chylous ascites. This fluid collects in the peritoneum either due to leaking lymphatics following trauma or due to obstruction of the lymphatic ducts.[1] Congenital chylous ascites remains the leading cause in pediatric age group and if a cause cannot be found after appropriate diagnostic work up, it is labelled as idiopathic. Congenital lymphatic malformations like cystic lymphangioma and chylolymphatic mesenteric cysts are the most common to cause chylous ascites.[2] Patients may present with a severe cachexic look with muscle wasting, pleural effusions, palpable abdominal masses or hernias of umbilical or inguinal region.[3] Initial management involves drainage of chylous fluid, respiratory support if needed, diet rich in medium chain triglycerides (MCT) with or without total parenteral nutrition (TPN) followed by surgical interventions in cases of failed conservative treatment.[4] Many children with congenital chylous ascites present with abdominal distension, abdominal mass or hernias. These patients are diagnosed in the pre-operative period while evaluating for the abdominal conditions. Rarely, in cases of mild ascites they are diagnosed intra-operatively while operating for inguinal hernias.[5] We have managed a similar case of congenital chylous ascites as surprize during hernia surgery and later it turned out to be cystic lymphangioma on further evaluation. This case report aims to highlight lymphovascular anomaly as the main etiological factor in majority of congenital chylous ascites in infants. Their individualized management strategies emphasizing on surgical resection and a detailed review of literature is discussed.

CASE

A 5 month old boy presented for right sided inguinal swelling. The swelling increased on coughing and crying and reduces on sleep. There was no history of fever vomiting and pain in the swelling. No feeding or respiratory difficulty was there. On examination there was a reducible, non-obstructed right inguinal hernia with palpable bowel loops as contents.

Both the testes were palpable separately and the scrotum was normal. Left side inguinal region was also normal. There was no abdominal distension and tenderness on examination. After baseline work up and pre-anaesthetic fitness, the child was taken up for hernia surgery. During surgery as soon as the hernial sac dissected and opened, there was a gush of milky white peritoneal fluid egressed from deep ring (**Figure 1a, 1b**). Around 100 ml of similar fluid was drained, sent for fluid analysis and herniotomy completed. The patient remained stable in the post-operative period with no obvious abdominal signs and tolerated orals well. Due to personal constraints, the parents took the patient home despite being counselled about the risk of serious complications of chylous ascites and the need for urgent evaluation. The patient returned for assessment after a few days." Ultrasonography was suggestive of moderate ascites with a 30x34x45 mm cystic lesion in the mesentery. Contrast CT abdomen findings were suggestive of a 32x30x48 mm sized mildly enhancing cystic lesion with internal calcifications in left paraumbilical region with ascites (**Figure 2a, 2b**). Chest X-ray was normal. The ascitic fluid (sent intra-operatively during herniotomy) analysis was consistent with chylous ascites (**Table**). Haematological and serum biochemistry parameters were within normal limits. Lymphangiography and lymphoscintigraphy studies were not done due to unavailability in nearby places. So the child was planned for surgical exploration with a working diagnosis of a cystic lesion of lymphovascular origin based on the radiological and laboratory findings. Intra-operatively there was chylous ascites of around 1200 ml with a ruptured cystic lesion of size 35x32x45 mm in the mesentery of small bowel at around 60 cm proximal to IC junction. There were spicules of calcifications in the wall of the cavity and copious amount of lymphatic fluid was leaking from the cyst wall (**Figure 2c**). The cystic lesion was resected along with the corresponding segment of bowel and end to end ileo-ileal anastomosis was done (**Figure 2d**). Post operatively the child remained stable. The abdominal drain output was initially around 200 ml (serous) per day which gradually decreased to less than 10 ml and then removed after 10 days. Histopathology was suggestive of dilated lymphatic channels and lymphoid cell aggregation along with collection of inflammatory cells and areas of hemorrhage diagnostic of cystic lymphangioma (**Figure 3a, 3b**). The child was on regular follow up after discharge from the hospital for the assessment of growth, appetite and abdominal symptoms. At one year follow up the child is symptom free and growing well.

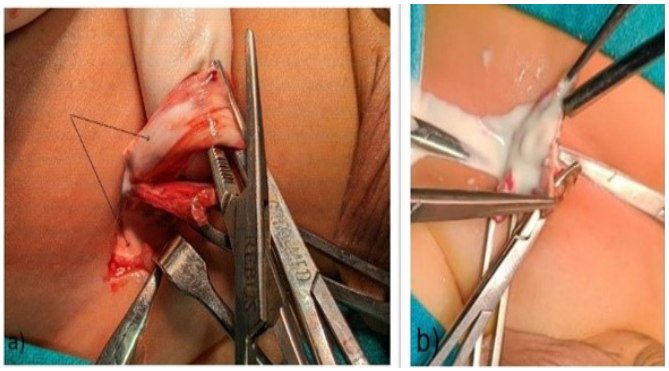

Figure 1. Intraoperative picture of chylous fluid: **a**) within and **b**) outside the hernia sac (black arrows).

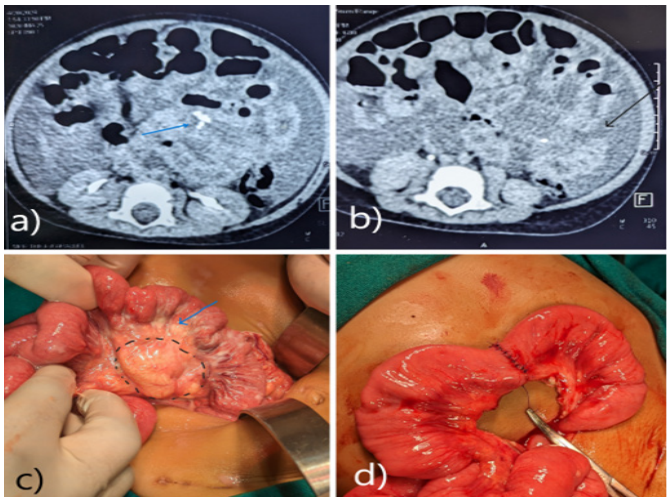

Figure 2. CT images and intraoperative photographs of the cystic lesion with chylous fluid: **a**) Axial CT cut with calcified mass in the mesentery (blue arrow) **b**) CT image with ascites (black arrow). **c**) cystic lymphatic mass in the mesentery and d) Bowel with mesenteric defect after resection and anastomosis.

Table. Peritoneal fluid analysis

Test name	Results	Units
Appearance	Milky white peritoneal fluid for analysis	
Triglycerides	9528.4	mg/dl
Cholesterol	119.44	mg/dl
Chylomicrons	Positive	Negative
Amylase	26.24	U/L
Microscopy	Lymphocytes	N/A
Cytology	Moderately cellular smear showing lymphocytes, neutrophils, mesothelial cells and histiocytes	N/A
ZN stain	Negative for acid fast bacilli	N/A
Culture & sensitivity	Negative for bacterial growth	

U/L: Units per liter, N/A: Not available, ZN stain: Ziehl-neelsen stain

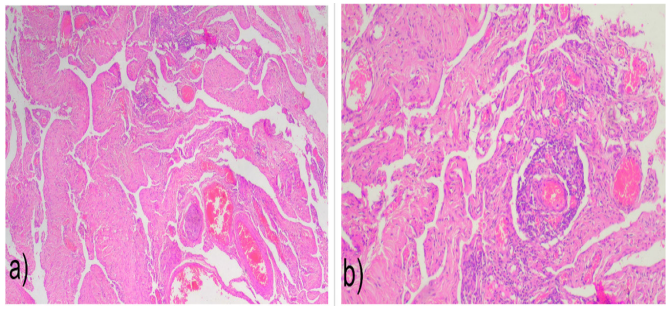

Figure 3. Histopathology (H & E stain) showing **a**) dilated lymphatic channels and cystic spaces with lymphoid aggregates and inflammatory cells (4X). **b**) Proteinaceous and hemorrhagic contents with dilated cystic spaces (10X).

DISCUSSION

Interstitial spaces of the body produce lymphatic fluid which transports nutrients, electrolytes, hormones and pathogen via a unidirectional valved lymphatic system. These vessels take lymph to the lymph nodes containing immune cells to filter the cellular waste, water, proteins and pathogens. Abdominal lymphatics absorb fat from bowel and take the lymph from abdominal organs. They join the lumbar lymphatics to form

the cisterna chyli and drain into venous circulation at the confluence between left subclavian vein and jugular vein. The lymphatic system is a one way drainage system which includes lymph, lymph vessels, lymphatic tissues and bone marrow. Any injury or obstruction to these ducts leads to chylous effusions or chylous ascites depending on the site of obstruction and this is the principle mechanism behind the chylous ascites formation.[6]

Chylous ascites has been reported since 17th century in various forms. Based on lymphangiography studies following causes have been identified.[6]

- Congenital lymphatic anomalies like lymphangioma or lymphatic hypoplasia.
- Cardiac causes such as cardiac failure, dilated cardiomyopathy and pericarditis.
- Infections like tuberculosis, Filariasis and mycobacterium avium.
- Inflammatory conditions like radiation, pancreatitis, retroperitoneal fibrosis and sarcoidosis.
- Gastrointestinal disorders like Malrotation and volvulus.
- Metabolic disorders like tyrosinemia and glycosylation disorders.
- Traumatic abdominal or thoracic injury
- Renal causes like nephrotic syndrome, liver cirrhosis and portal hypertension.
- Post-operative conditions like congenital diaphragmatic hernia, nephrectomy and fundoplication.
- Neoplastic causes like lymphangiomyomatosis and Kaposi's sarcoma.
- Drugs like calcium channel blockers
- Other causes like incarcerated inguinal hernia and obturator hernia.

Chylous ascites presents mostly as vague pain and abdominal distension in more than 90% of the patients. In infants it can present as umbilical or inguinal hernias as in the index case, though very rare. Post-surgical patients with duct injury presents as acute respiratory distress, abdominal distension, edema, cachexia, muscle wasting and effusions.[7]

Laboratory analysis of the chylous fluid is the most confirmatory investigation for the diagnosis of chylous ascites. The fluid is analysed for the cell count, gram stain, triglyceride levels, total protein, albumin, bacteria, culture, lactate dehydrogenase (LDH), amylase and glucose.[8] Chylous fluid has triglyceride levels higher than 110 mg/dl as mentioned by a few authors however, they are mostly above 200 mg/dl.[7] Our patient had triglycerides levels near 10,000 mg/dl (Table), suggesting the fluid to be chyle only.

Contrast CT scan can easily pick up and localize lymphatic mass/malformations, type of fluid (blood/ascites), thoracic duct injury and other calcified lesions if any.[7] We have done a contrast CT abdomen and found an enhancing lesion with internal calcifications in mesentery and gross ascites.

Lymphangiography is the gold-standard diagnostic modality for assessing lymphatic anatomy, locating sites of leakage or obstruction, and evaluating the retroperitoneal lymph nodes and thoracic duct. However, because it is invasive and carries risks such as hypersensitivity reactions, fat embolism, and tissue necrosis, it is increasingly being replaced by non-invasive investigations.[9] In infants with congenital lymphatic malformations, it helps distinguish between lymphatic hypoplasia, aplasia, and localize leakage. The study involves subcutaneous injection of a radiolabeled tracer (commonly Tc-99m sulphur colloid), followed by serial imaging to trace lymphatic drainage. When combined with MR lymphangiography, lymphoscintigraphy offers complementary physiologic information, aiding in accurate diagnosis and guiding surgical or interventional management. It is also used in the post-operative period to assess the effectiveness of a lymphatic bypass or thoracic duct ligation. Lymphoscintigraphy is used when lymphangiography is contraindicated to assess the lymphatic drainage and functional assessment. Although it has no specific contraindications but, it is still not used frequently due to technical challenges involved in small children.[10]

Diagnostic laparoscopy is reserved for patients with no definitive cause identified and suspicion of malignancy and tuberculosis. In post-operative patients sometimes exploratory laparotomy is required to diagnose and treat the underlying cause. It is also combined with lymphangiography intra-operatively to properly delineate lymphatic anatomy.[8]

Management of chylous ascites involves multimodality treatment involving nutritional specialist, pharmacotherapies and surgical management. In patients with an identifiable cause, treatment of primary condition is the mainstay of treatment. All the medical management strategies are based on decreasing chyle flow by bowel rest, TPN, replacing electrolytes and micronutrients. This resolves chylous ascites in majority of the patients.[11]

Dietary modifications include high protein diet enriched with MCT and restriction of long chain triglycerides (LCT). MCTs are absorbed from intestines and transported directly into liver as free fatty acids and glycerol, preventing both production and flow of chyle. Patients not responding to these measures require bowel rest and TPN.[10] Along with TPN, Somatostatin analogue octreotide has been found very effective in rapidly reducing the chylous ascites, probably by inhibition of lymph secretion from lymphatic vessels.[12] Transjugular intrahepatic portosystemic shunt (TIPS) is used in cirrhotic patients with end stage liver disease however, it has not been approved in children.[13] Peritoneovenous shunts were used in past as a rescue therapy for patients not responding to medical management and not fit for surgery.[14] Angiographic embolization is also used for resistant chylous ascites as an effective method.[15] Chylous ascites of congenital causes, neoplastic etiology or post-operative origin may need

surgical exploration however, operative interventions are not without complications.

There are a few published reports in literature on chylous ascites in infants and their management. Causes of chylous ascites in infants are mainly congenital lymphovascular disorders, incarcerated hernias, intestinal malrotation with volvulus, liver disease and thoracic duct atresia. Sometimes the ascites presents as a diagnostic surprize during hernia surgery in a previously asymptomatic child as in index case.[6]

Alnee et al.[1] in 2011, managed a similar case of chylous ascites presented with abdominal distension and bilateral inguinal hernias by repeated paracentesis and conservative management.

In 2021, Dreuning et al.[6] concluded that the chylous ascites in an infant with inguinal hernia can be due to mechanical compression of lymphatics during repeated incarceration and attempted manual reduction of contents if no other cause found on further work up. This can be managed very well with MCT diet and conservative approach.

Siddiqui et al.[16] also managed similar case with conservative approach in an infant with chylous ascites presenting with bilateral inguinal and umbilical hernia. However, they could managed to get lymphoscintigraphy done and identified the cause of chylous ascites as leak from retroperitoneal lymphatics.

Albaghdady et al.[17] published a series of four infants with chylous ascites managed by surgical interventions combined with medical management. Surgical interventions included resection of chylolymphatic cyst, resection of omental cyst and placement of peritoneovenous shunts.

Presence of calcification is another significant finding seen rarely. The differential diagnoses for calcified cystic lesion of mesentery include teratoma, meconium peritonitis cyst, dermoid cyst, hydatid cyst, pseudocyst and calcified hematoma. To the best of our knowledge only four cases of calcification in cystic lymphangioma are reported and they were managed by surgical excision. Exact etiology for the calcifications is not clear however, there is a possibility that presence of the lesion from the intrauterine life and chronic inflammation could have led to calcium deposition in the walls of the lesion.[18]

It is evident from above discussion that most of the neonates with chylous ascites responds well to medical management and very rarely surgical exploration is needed. Those who need operative treatment are the cases of either active lymphatic leak or radiologically proven lymphovascular malformations. Resection of congenital mass and involved part of bowel as in index case, closure of chyle leak and repair of retroperitoneal lymphatic fistulas are few surgical procedures used with good outcomes. Sometimes other modalities like angiographic embolization, fibrin glue peritoneovenous shunts and use of etilefrine might need to be utilized in failed surgical explorations.[9] However chylous ascites diagnosed unexpectedly during hernia surgery in an asymptomatic child is very rare. In our patient surgical intervention combined with medical management were successful in controlling the chylous ascites effectively over a period of two weeks. The uniqueness of this case lies in the rarity of the diagnosis and its incidental detection during a commonly performed procedure in an infant.

CONCLUSION

This case illustrates the importance of intraoperative awareness and rapid diagnostic evaluation when faced with atypical findings during routine procedures like herniotomy. Although chylous ascites is often associated with serious underlying conditions, it may occasionally present in otherwise asymptomatic infants, as in our case. Conservative management remains the first-line approach, with favourable outcomes in idiopathic or postoperative cases. A few might need further evaluation and surgical intervention. In infants with ruptured cystic lymphangioma, complete resection of the lesion with restoration of intestinal continuity provides the best therapeutic results and is associated with good postoperative outcomes. Reporting such rare presentations enhances our understanding and preparedness for unexpected intraoperative challenges in pediatric surgical care.

ETHICAL DECLARATIONS

Informed Consent

Informed consent was obtained from the legal guardians of the pediatric patient(s) described in this report. Where developmentally appropriate, assent was also sought from the child. The inclusion of vulnerable populations in this study adhered to national and international ethical guidelines. Extra care was taken to ensure voluntary participation, understanding, and protection of participant dignity and autonomy.

Peer Review Process

This manuscript was subject to external peer review.

Conflict of Interest

The authors declare no conflicts of interest related to this study.

Financial Disclosure

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Author Contributions

The author is solely responsible for the conception, data collection, analysis, and writing of this manuscript.

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