Case Report

Refractory Congenital Chylous Ascites: Role of Fibrin Glue in its Management

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Chylous ascites is the accumulation of triglyceride-rich lymph in the abdomen. Its occurrence during the infantile period is quite rare. Congenital chylous ascites (CCA) is one, which occurs in children <3 months of age, due to maldevelopment of the lymphatic system. There is no clearly defined treatment protocol for CCA; however, the use of medium-chain triglycerides (MCT)-based diet or total parenteral nutrition (TPN) with octreotide has been successful. Failure of conservative management, however, leads to surgical exploration to deal with those leaking lymphatics. In our case, we had initially given a trial of managing the child with MCT-based diet followed by a TPN along with octreotide. However, the failure of both leads us to operate the child during which we incorporated the use of fibrin glue over the leaking mesenteric lymphatic vesicles, which ultimately led to the resolution of the chylous ascites.

Keywords: Congenital chylous ascites, fibrin glue, neonatal chylous ascites

INTRODUCTION

Congenital chylous ascites (CCA) is a rare entity, which presents as an accumulation of chyle in the abdominal cavity of children <3 months of age. The composition of chyle is rich in fat globules with triglycerides valuing at twice or thrice than that of the serum. It occurs due to leaking from lymphatics either due to obstruction, trauma, or congenital maldevelopment. Management usually consists of medium-chain triglycerides (MCT)-based diet or total parenteral nutrition (TPN), and the addition of octreotide, which is a somatostatin analog has been reported to be immensely useful. Surgical exploration is reserved for those cases in whom conservative management has failed.

CASE REPORT

An 8-week-old male infant born from a nonconsanguineous marriage by normal vaginal delivery was admitted with complaints of rapidly increasing abdominal distension since day 5 of life. On examination, the patient was dyspneic, and the abdomen was grossly distended with a girth of 46 cm. Ultrasound abdomen was suggestive of gross ascites [Figure 1a]. Abdominal paracentesis was done, which suggested chylous ascites. The milky fluid had triglyceride levels of >8000 mg/L, sugar 42 mg/dl, and protein 3.8 g/dl. An MCT-based diet along with octreotide infusion at 1 μg/kg/h was started and continued for 2 weeks, but the ascites persisted. Computed tomography (CT) scan was suggestive of gross ascites, and no lymphatic malformation/cyst was identified [Figure 1b and c]. Lymphosynctigraphy was planned, but was not carried forward due to technical difficulties. Repeated paracentesis was required to relieve respiratory distress due to abdominal distension. TPN was started while escalating octreotide to 3 μg/kg/h, which dramatically reduced his abdominal girth from 46 cm to 36 cm in 28 days. However, ascites recurred on starting oral feeds. Magnetic resonance imaging (MRI) was done, which suggested the recurrence of chylous ascites, but no lesion was identified. Surgical exploration was planned, and the child was fed fat-rich diet mixed with Sudan dye 6 h before the surgery, as we believed...
that it would further help us in delineating the leaking points during surgery.

On diagnostic laparoscopy, ascitic fluid was seen [Figure 1d], but no remarkable lesions were identified, so it was converted to open surgery. Intraoperatively, multiple prominent lymphatic vesicles [Figure 2a] were noted at the base of the mesentery in the ileocecal region over which fibrin glue was applied [Figure 2b]. There was no staining of lymphatics by the lipophilic dye. Postoperatively, TPN along with octreotide at 1 μg/kg/h was continued for 8 more days. Oral diet was slowly initiated until the child was able to tolerate full breastfeed. The abdominal girth remained stable at 35 cm until the day of discharge after 2 months of hospital admission.

The child is presently in close follow-up with an abdominal ultrasound planned every 2 weeks for a month followed by once a month for the next 3 months.

**DISCUSSION**

CCA is defined as one, which occurs in a child <3 months of age.[1] Although it is an uncommon condition with a reported incidence of 1:20,000–1:187000, more than half of them present in infants.[3,4] The majority of cases occur due to some lymphatic malformation, which can be atresia or stenosis of lymphatics, lymphangiomatosis, and chylous cysts. About 30% of cases are idiopathic, in which “leaky lymphatics” due to delayed maturation of lacteals lead to CCA.[2‑4] Other causes include external compression of lymphatics due to malrotation, intussusception, malignancy, or trauma during surgery, accidents, or child abuse.[2] The patient usually presents with abdominal distension with or without respiratory distresses our patient and respiratory distress calls for therapeutic tap. Due to the constant loss of proteins and lymphocytes, chylous ascites have other dreadful complications too, which are related to mechanical, nutritional, and immunological factors.

The ascitic fluid is usually milky although it might be straw colored in some patients not fed enterally. The primary diagnosis is through paracentesis and analysis of the fluid, which will have high triglycerides levels and chylomicrons. Imaging studies such as ultrasonography, CT, and MRI, which can detect malformations. The imaging study of choice is scintigraphy, which evaluates the patency of lymphatics and identifies the site of leakage,[2,3] but it may be difficult to perform in neonates and small infants.

Most CCA have been managed conservatively, which would commonly consist of keeping the child off of breast milk and initiating an MCT-based diet or on full TPN and octreotide.[2] The conservative treatment should continue for 4–8 weeks.[2‑3] The success rate of conservative treatment is around 60%–100%.[5] In our patient, we continue conservative treatment initially by MCT-based diet for 2 weeks and TPN with octreotide infusion for 4 weeks. However, the ascites reappeared as soon as we fed the patient orally. The failure of conservative management warrants a surgical exploration, usually consisting of resection of the visible localized anomaly or ligation of the leaking lymphatic vessels, but only 58% of patients with CCA may have a surgically treatable lesion.[3] The preoperative localization of leaking sites is indispensable and other than imaging and lymphoscintigraphy, administration of lipophilic dye like Sudan black 6 h before the surgery may help to direct visualization of leaking sites.[2‑3]

Even if it does not stain lymphatics, as in our case, the lipid-rich diet may make the lymphatics prominent.

The surgical management includes exploration of all the major lymphatics and suturing the leak point, excision of the lymphatic cyst, omentum, or another identifiable

![Figure 1](image1.png)

**Figure 1:** (a) Preoperative image showing tense ascites; (b) *Computed tomography abdomen showing gross ascites in axial and (c) *Computed tomography abdomen showing gross ascites in coronal plane (d) *Chylous ascites during diagnostic laparoscopy

![Figure 2](image2.png)

**Figure 2:** (a) *Mesenteric lymphatic vesicles; (b) *Fibrin glue applied over the mesenteric lymphatics
lesion.[2,3] If there is no obvious cause, we should explore the root of mesentery by kocherisation of the duodenum and identify the cisterna chyli, which lies between the aorta and inferior vena cava.[2,6]

When the identification and ligation of specific leak sites are not possible as in leaky lymphatics and small neonatal lymphatics, fibrin glue is used to supplement the ligature, absorbable mesh, or as exclusive topical application to seal the diffuse leakage.[2,3]

In our patient since the ligation of lymphatics over the surface of the mesentery was not possible, we decided to seal them with fibrin glue, which eventually stopped the leaking of chyle from lymphatics.

The peritoneovenous shunt is indicated when both nonoperative and operative measures fail to resolve CCA, which is associated with a high risk of failure and complications.[2,3,5]

We could not identify the leakage site on laparoscopy although it is reported to be useful in the identification of leakage, as it provides a magnified view of the natural state of the peritoneal cavity and helps in ligation of the lymphatic vessels.[3,5]

Fibrin sealant, also referred to as “fibrin glue” or “fibrin tissue adhesive,” is a surgical hemostatic agent derived from plasma coagulation proteins. The clinical use of fibrin was first reported in 1909 by Bergel and in 1938 purified thrombin became available for commercial use. However, the combination of both was first used in 1944 to enhance the adhesion of skin grafts in soldiers with severe burn injuries.[7] Fibrin sealant, which is now available as a two-component material, consisting of fibrinogen and thrombin. In our case, we used EVICEL®: Ethicon Inc. (Fibrin Sealant [Human]), which consists of two vials of 1 ml each of thrombin and fibrinogen delivered through a triple lumen tip. The two-component fibrin sealant is usually applied through a double-barreled syringe system, which allows the simultaneous application of equal volumes of the fibrinogen and thrombin through a blunt-ended needle or spray tip. It is stored in frozen vials at −18°C or colder for up to 2 years and thawed before the use at 20°C to 25°C (room temperature) within 1 h; or at 37°C (warm water bath): vials thaw within 10 min and must not be left at this temperature for longer than 10 min. The temperature must not exceed 37°C. As an approximate guide, a layer of 1-mm thickness is produced by spraying EVICEL® is appropriate.

**CONCLUSION**

Although the diagnosis of CCA is straight forward, but the management is very demanding and bears an unprecedented course. Although most of the cases are managed conservatively, the surgery may be required for refractory cases. The prolonged course of TPN and octreotide makes it difficult to dispense in resource-limited settings and enduring to patients, parents, and treating team. The localization and ligation of leakage sites are difficult, especially in newborns and young infants due to small size lymphatics. The use of fibrin glue to seal the lymphatics is quite promising, especially in cases where one cannot identify any specific site of leakage.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal patient identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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