Dual imaging lymphangiography guided treatment of infantile chylothorax

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ABSTRACT

Chylothorax is a potentially fatal postoperative complication of neck, thoracic, and abdominal surgery in children. We report the case of a 3-month-old infant who developed persistent chylothorax with respiratory insufficiency successfully managed using a microsurgical technique and intraoperative embolotherapy. This was achieved using a combination of intermittent digital X rays and live near-infrared fluorescence imaging we have termed "dual imaging lymphangiography" to guide therapy in real time. The chylothorax resolved and the patient returned to normal diet without recurrence. This microsurgical approach with dual imaging lymphangiography provides a useful tool for intraoperative visualization and treatment of complicated chylothoraces. (J Vasc Surg Cases Innov Tech 2021;7:492-5.)

Keywords: Infantile chylothorax; Chylous leak; Chylous ascites; Lymphangiography; Microsurgery techniques

Postoperative chylothorax is a potentially life-threatening complication of congenital heart disease surgery in children that affects 2% to 9% of cases.[1-3] The continuous loss of chyle can result in immunologic compromise, malnutrition, metabolic disorders, fluid and electrolyte imbalance, and death in 6% to 18% of patients.[1-6] Management options include conservative therapy, embolization therapy, and surgery; however, consensus on optimal treatment options remains disputed particularly in complex persistent cases.

We present a case of chylothorax after congenital heart disease surgery that persisted after conventional management. This was successfully treated using microsurgical techniques with a combination of intermittent digital X rays and live near-infrared (NIR) imaging we have termed "dual imaging lymphangiography" (DIL) to guide therapy in real time.

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CASE REPORT

A 3-month-old infant developed right-sided chylothorax with respiratory insufficiency 3 days after pulmonary artery banding and patent ductus arteriosus ligation surgery for congenital double-outlet right ventricle with patent ductus arteriosus. Conservative therapy with median chain triglyceride (MCT) diet and octreotide were commenced, and a chest drain was inserted. However, 3 weeks later the chylothorax and reduced oxygen saturation remained unimproved and the child was referred to our department for further evaluation and treatment.

First, we performed lymphoscintigraphy and single photon emission computed tomography, confirming lymphatic leakage into the right thoracic cavity and identifying the approximate location of the leakage at the height of the 11th thoracic vertebra (Fig 1, *A* and *B*; Supplementary Video 1, online only). Although these modalities were not suitable for identifying finer leaks due to their low resolution, they were useful for screening for leakages in other sites such as ascites both preoperatively (Fig 1, *A* and *B*) and postoperatively (Fig 1, *C* and *D*).

Single lung ventilation was then commenced, and a right thoracotomy performed to access the thoracic cavity for detection and ligation of leakage sites. Using an operating microscope (OPMI Pentero 900; ZEISS Japan, Tokyo, Japan), the thoracic duct (TD) (1.5-2 mm in diameter) and its branches (0.5-0.7 mm in diameter) were identified and the leakage site was identified at the inferior TD (Fig 2, *A* and *B*). The TD and its branches were then ligated on the caudal side by clipping using a microsurgical technique.

Postoperatively, the child was kept on an MCT diet and the amount of pleural effusion gradually decreased for 10 days. However, on introduction of normal milk, the chylothorax returned. Repeat lymphoscintigraphy and single photon emission computed tomography were performed and lymph leakage was found at the height of the diaphragm on the inferior side of the ligated TD (Fig 1, *C* and *D*).

The child was returned to theater for lymph vessel ligation and embolization. A transverse abdominal incision was made, and the lymphatic vessels were identified using indocyanine green

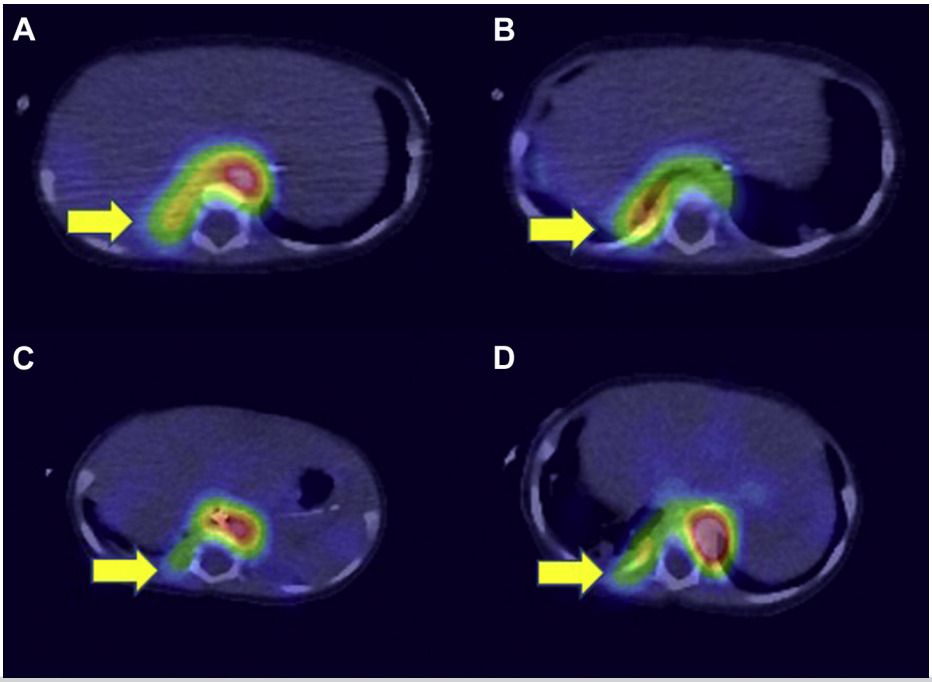

Fig 1. Single photon emission computer tomography (SPECT) lymphangiography images showing lymphatic flow in the chest region. Serial preoperative images **(A and B)** show lymphatic leakage into the right chest cavity from the thoracic duct (TD) detected as with lymphoscintigraphy at the height of the 11th thoracic vertebra (*yellow arrow*). Repeat serial SPECT images after thoracic duct (TD) ligation **(C and D)** show persistent lymphatic leakage inferior to the TD ligation site.

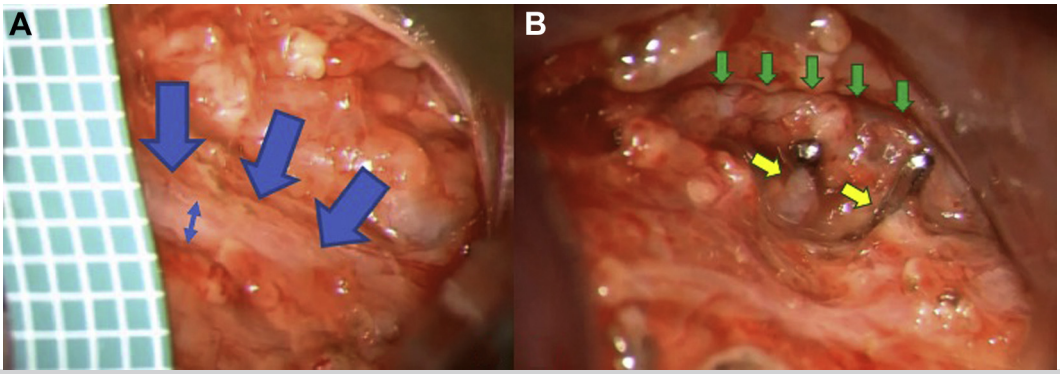

Fig 2. Microsurgical thoracic duct (TD) identification and dissection. **A,** The TD (*blue arrow*) 1.5-2 mm in diameter (*blue two-direction arrow*) and its branches (0.5-0.7 mm in diameter) were identified. **B,** The TD (*green arrow*) and its branches (*yellow arrow*) were then ligated on the caudal side by clipping.

(ICG; 0.2 mL, 2.5 mg/mL, Diagnogreen; Daiichi Sankyo, Japan) lymphangiography and operating microscope NIR imaging. A suitable intra-abdominal lymph node was then selected, and lipiodol (1 mL, ethyl ester of iodinated poppy-seed oil fatty acid; Guerbet Japan, Tokyo, Japan) and ICG (1 mL, 25 mg/mL) in the ratio 1:1 were injected at the same time into the lymph node under the microscope (Supplementary Video 2, online only), observing the flow of the agent using NIR imaging as we previously reported.[7] In addition, portable digital X ray images (FUJIFILM DR CALNEO Go; Fujifilm, Tokyo, Japan) were taken intermittently, viewed with a high-resolution tablet (TOUGHPAD 4K; Panasonic, Osaka, Japan) to confirm the lipiodol embolization of the thoracic lymphatics (Fig 3, *A* and *B*).

After surgery, feeds were withheld and then an MCT diet was started after 2 weeks. A month later, breast milk was commenced, the chest drain was removed, and the patient was discharged. Review at 6 months after surgery revealed no recurrence with the child in good health and normal growth, and finally the patient underwent double-outlet right ventricle repair and pulmonary artery debanding at 14 months of age, without any recurrence of chylothorax (Fig 4, *A* and *B*). The patient has provided consent for this case report.

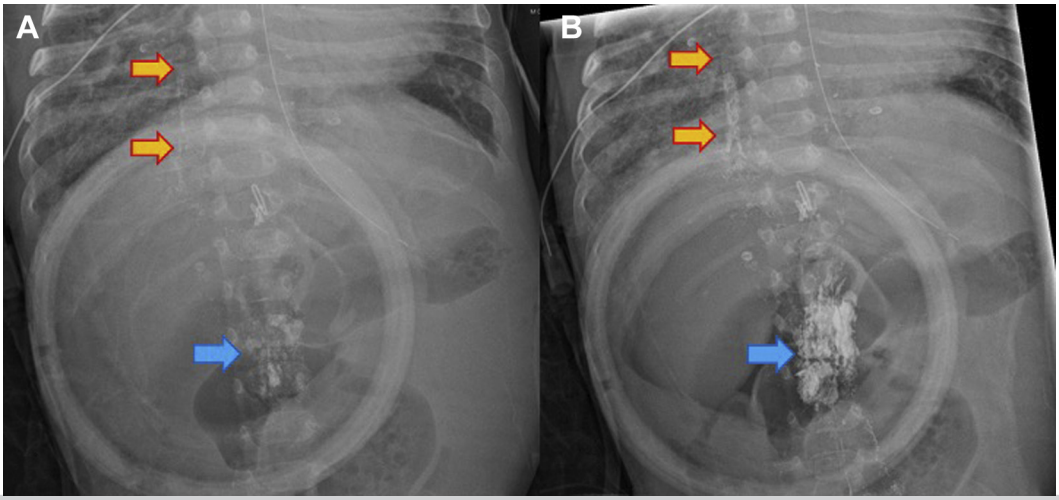

Fig 3. Serial intraoperative intermittent digital X ray lymphangiography images after the injection of a mixture of lipiodol and ICG into an intra-abdominal lymph node (*blue arrow*), which confirmed that it was flowing into lymphatic vessels (*orange arrow*). X ray was performed three times: **(A)** the first image and **(B)** the third image.

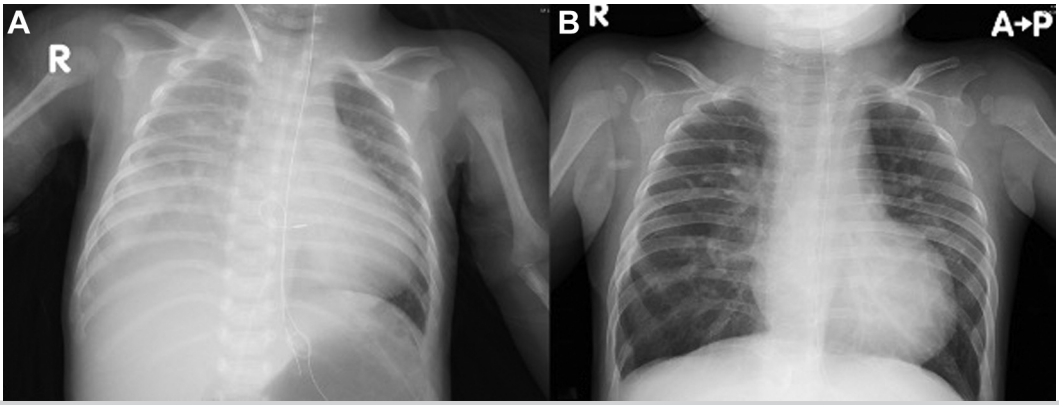

Fig 4. Pretreatment chest X ray **(A)** shows the features of right-sided chylothorax preoperatively, and postoperative chest X ray 6 months after the final surgery **(B)** shows no recurrence of chylothorax.

DISCUSSION

Chylothorax remains an important source of morbidity in pediatric cardiothoracic surgery. It is associated with prolonged hospital stay, delayed ventilator withdrawal, higher cost of treatment, and increased mortality.[2,3] The incidence of chylothorax after cardiac surgery significantly increased from 2% in 2004 to 3.7% in 2011.[3] Both incidence and mortality are significantly higher in neonates and infants, and decrease with growing age.[3]

At present, there is no consensus on the best treatment of postoperative chylothorax in children. This is partly attributable to the diverse etiology of chylothorax in post congenital heart disease surgery compounded by the limitations in diagnostic imaging in defining lymphatic disease in young children. A recent study by Savla et al[8] using dynamic contrast-enhanced magnetic resonance lymphangiography found that nontraumatic chylothorax due to abnormal pulmonary lymphatic perfusion and complex central lymphatic anatomy accounted for the majority of cases, whereas direct traumatic leakage was found in only 8% of postoperative chylothorax in patients with congenital heart disease. Nevertheless, conservative treatment is successful in 80% of patients, whereas other treatment modalities such as TD ligation and embolization by percutaneous interventional radiology are often reserved for persistent cases.[5,9] When suitable lymphatic draining vessels are present, lymphaticovenous bypass is also used to treat infantile chylothorax.[9]

In our patient, we were able to dissect and identify the leakage sites on the TD and its tributaries and perform selective ligation using the operating microscope and supermicrosurgical technique. Traditionally, detection and ligation of the TD has been performed without a

microscope since Lampson reported first TD ligation for chylothorax in 1948.[10] When used in the treatment of chylothorax in young infants, this can be a difficult procedure. In these patients, the use of the operating microscope equipped with an NIR camera makes detection and dissection of lymphatic channels considerably easier. Blue dyes may also be used as an affordable alternative to microscopical NIR imaging, but they are limited to visualization of only surface lymphatics.

For the second operation, a transabdominal approach was opted to avoid the thoracic adhesions and postoperative atelectasis. This also allowed access to the larger abdominal lymphatic vasculature and nodes for microsurgical treatment. We used a mixture of lipiodol with ICG for embolization. This allowed us to visually confirm the correct flow of the lipiodol embolization agent from the nodes into the lymphatic vessels in real time using operating microscope NIR imaging. NIR provides high-resolution, safe, relatively low-cost imaging with no ionizing radiation exposure. However, in contrast to X rays, it is limited by low depth of tissue penetration. Therefore, portable digital X ray was used to confirm the embolus site in the upper abdomen and chest after lipiodol injection because of its higher resolution compared with fluoroscopic images, making it possible to identify the deeper lymphatic vasculature. By injecting intermittently and photographing several times, it was also possible to confirm the arrival point of the contrast agent while avoiding continuous exposure. This microsurgical approach with DIL enhanced the secure identification of lymphatic channels and surgical treatment with minimal radiation exposure. Furthermore, it enabled confirmation of the location of the embolotherapy agent at higher resolution without moving the patient. Lastly, DIL can be performed without the need for a hybrid operating room few hospitals have ready access; thus DIL may be useful in such centers.

CONCLUSIONS

The microsurgical approach with DIL provides a useful, relatively inexpensive, tool for the treatment of complicated chylothorax in infants.

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