

Chylovenous bypass for mesenteric lymphangiomas: A case report

We thank authors Miyazaki et al for their thoughtful comments. Indocyanine green (ICG) lymphography is a powerful tool for identification of both normal lymphatic vessels and any aberrancies such as drainage or dermal backflow, which is characterized by diffuse infiltration of lymph.^{1,2} It is even, in certain settings, a superior diagnostic imaging method compared to other modalities such as lymphoscintigraphy.³ Typically, when mapping lymphatics with ICG, the ICG is injected distally, such as in the webspace of the hands or feet. For retroperitoneal lymphangiomas (RL), ICG was directed more locally with injection at the distal site of the RL to evaluate the chyle drainage direction before microsurgical chylovenous bypass and to confirm patency of the anastomosis.⁴

Retroperitoneal lymphangiomas—a malformation of the lymphatic system in the retroperitoneal space—is a rare form of primary lymphedema.^{4,5} While the retroperitoneal lymphatic malformation itself is benign, the concomitant symptoms that result from the lymphatic/chylous leakages are not. In our study, we found most of the chylous ascites were a result of RL and could be circumvented by chylovenous bypass.⁴ In the case of chyle leakage secondary to mesenteric, rather than retroperitoneal, lymphangiomas, injection of ICG into the subserosal layer of the distal mesentery as Miyazaki et al suggested in their letter was effective in identifying lymphatic defects of interest, as seen in our case report of a 3-year-old patient with chylous ascites and primary extremity lymphedema.

The 3-year-old patient presented with abdominal distention, chronic diarrhea, bilateral hydrocele, and right lower extremity lymphedema. Lymphoscintigraphy revealed a partial lymphatic obstruction in the right distal lower extremity with bilateral enlarged and atypical inguinal lymph nodes. Ultrasound Doppler and computer tomography demonstrated ascites in the perihepatic and perisplenic spaces, the paracolic gutter, and the pelvis. Findings of dilated, tortuous lymphatic vessels in the lower chest, abdominal para-aortic, and iliac regions in addition to mesenteric edema and prominent para-aortic, iliac, and inguinal lymph nodes were seen on magnetic resonance imaging (MRI).

Despite the bilateral herniorrhaphy and hydrocelectomy with right orchiopexy performed in October 2017, the patient continued to have persistent scrotal swelling (Figure 1A). The refractory nature of the patient's disease and malnutrition made surgical intervention indicated.⁶ An exploratory laparotomy was arranged in March 2019. No chyle leakage was found from the retroperitoneum, but 100 mL of accumulated chyle inside the mesentery and omentum was noted and

aspirated (Figure 1C). Intraoperatively, a lymphangioma-like lesion was detected on the mesentery and omentum. ICG injection was utilized to confirm extravasation from the lesion. Once confirmed that the chyle originated from the mesenteric and omental lesion, a 1 mm in diameter, 12 cm in length segment of the left deep inferior epigastric vein (DIEV) was dissected in the caudal to cephalic direction (Figure 1D) and transpositioned to anastomose to the omentum in a side-to-end fashion using 11-0 Nylon sutures. The chylovenous anastomosis was confirmed to be patent (Figure 1E), with chyle successfully draining into the DIEV (Figure 1E). Within 3 days postoperatively, the scrotal swelling subsided (Figure 1B). The patient was placed on a nonfat diet for 1-month postoperatively⁷ before transitioning back to a normal diet. At the 15-month postoperative follow-up visit, the patient was tolerating a regular diet without signs or symptoms of recurrence of chylous ascites.

Operatively, the primary concerns were how to correct the chylous leakage and how to identify the source. In this case, direct visualization accompanied by secondary ICG-based confirmation localized the chyle origin to the diffuse mesentery of the intestine and related omentum. Symptomatically, the chylovenous bypass resolved the chylous ascites and downstream hydroceles, although it did not cure the direct etiology of the chylous ascites, which was the mesenteric lymphangioma itself. In such cases where the diagnosis of mesenteric lymphangiomas is based on intraoperative findings and imaging, ICG proves invaluable as a tool to characterize lymphatic drainage and likely can be applied in other instances where advanced preoperative imaging and direct intraoperative visualization are not entirely diagnostic or have pitfalls.⁸ However, lymphangiomas and its pathophysiology, along with related diseases, still require further inquiry. Lymphangiomas is a disease where lymph or chyle accumulation result from the overgrowth of lymphatic tissue or vessels.^{1,5} Similarly, lymphangiectasia, such as primary intestinal lymphangiectasia (Waldmann's disease), is another rare disorder with lymph leakage that is the result of dilated lacteals, and could be another disease state where ICG and chylovenous bypass could prove useful.^{9,10} Methods to diagnose, identify, and treat lymphatic diseases benign in nature but with harmful secondary effects continue to necessitate investigation.

To the author's best knowledge, this is the first case report of mesenteric lymphangiomas that was diagnosed by intraoperative findings and ICG lymphography and successfully treated by a chylovenous bypass.